Indonesian Journal of Medicine (2022), 07(03): 289-297
Masters Program in Public Health, Universitas Sebelas Maret

Case Report

Atypical Spinal Tuberculosis of Upper Thoracic Spine: A Rare Case Report

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ABSTRACT

Background: Tuberculous infection is one of the most common pulmonary infection in developing countries, including Indonesia. Spinal tuberculosis is one of the most common extrapulmonary tuberculosis manifestation, accounting for 5% of all extrapulmonary spread, and 50% of all osteoarticular involvement. The aim of this study is to evaluate and how diagnosis and treatment to prevent over or under-diagnosis and explain about excellent management without any late complication further.
Case Presentation: A 20-year-old male with a history of weakness of both of his leg for 20 days prior to admission. From these examinations, patient was then diagnosed with pathological fracture 2nd thoracic vertebral body suspected due to atypical spinal tuberculosis. Patient was then undergone surgical debridement, decompression, stabilization and fusion.
Results: Patient was then undergone surgical debridement, decompression stabilization fusion. During the exposure at level C6 through Th4, no abscess was found. Transpedicular debridement at level Th2 was then done after the insertion of pedicle screws at level C6-Th1 and Th3-Th4, and again no abscess was present, however, a granulomatous mass was present on the location of 2nd thoracic vertebral body, located on the anterior right side of the spinal cord outside the dura mater.
Conclusion: Atypical presentation of spinal tuberculosis with the sole clinical manifestation of neurological deficit below affected level. The definitive treatment such as surgical debridement, stabilization and fusion must be consider.

Keywords: spinal, infection, tuberculosis, young patient.

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Cite this as:
Wiguna IGLNAA, Wondany RP, Arimbawa IBG (2022). Atypical Spinal Tuberculosis of Upper Thoracic Spine: A Rare Case Report. Indones J Med. 07(03): 289-297. https://doi.org/10.26911/theijmed.2022.07.03.05.

BACKGROUND

Tuberculous infection is one of the most common pulmonary infection in developing countries, including Indonesia (Gede et al., 2008). Its extrapulmonary manifestation includes abdominal cavity, lymph node, genitourinary tract, skin, and osteoarticular tissues (Houston and Macallan, 2014). Spinal tuberculosis is one of the most common extrapulmonary tuberculosis manifestation, accounting for 5% of all extrapulmonary spread, and 50% of all osteoarticular involvement (Gautam et al., 2005). Lower thoracic and lumbar verte-

brae are the most commonly involved on this disease, which consists of 50% and 45% of all cases, respectively (Garg and Somvanshi, 2011). Three major clinical features of the spinal tuberculosis are: back pain, cold abscesses, neurologic deficit, and kyphotic deformity of the spine, which occurs in 90-100%, 70%, 10-20%, and 3% of all patients, respectively (Cormican, 2006). The management consists of surgical debridement and evacuation of the cold abscess, followed with vertebral stabilization and fusion as reported to restore spinal stability (Turgut, 2001).

This paper reports an unusual case of spinal tuberculosis involving 2nd thoracic vertebral body with absence on most of the major clinical features with no cold abscess formation and normal inflamematory markers.

CASE PRESENTATION

A 20-year-old male Balinese with a history of weakness of both of his leg for 20 days prior to admission. The weakness was initially felt around the hip area, and gradually spread to both of his legs around 10 days prior to admission. The weakness was accompanied with tingling sensation at the level of his chest down to tip of his toes. After the weakness has progressed, patient cannot walk by his own and mobilize using wheelchair. No micturition and defecation problem were experienced by the patient. No history of prolonged fever, chronic cough, weight loss, night sweat, and night pain. The medical report illness and family history were not found in this patient. Patient as an undergraduate student in Bali.

Patient has gone to a neurologist and received an injection and underwent a Magnetic Resonance Imaging (MRI) scan for his spine. After examining the MRI result, the neurologist referred the patient to Orthopaedic polyclinic for further management. Upon initial evaluation on the Orthopaedic polyclinic, there was no gross deformity seen on the whole spine, and no accompanying midline tenderness. Hypoesthesia at level Th3 was present, accompanied with paraparesis with the motoric strength around 3 on both legs using Medical Research Council (MRC) muscle strength grading system (3 for hip flexors, toe extensors and ankle plantarflexors; 4 for knee extensors; 2 for ankle dorsiflexors), which is classified as American Spinal Cord Injury Association (ASIA) neurologic function class C. Pathological reflexes were positive on both legs.

RESULTS

On the initial X-Ray examination, there was slight compression on 2nd thoracic vertebral body without any rarefaction on it. The MRI examination has shown severe compression fracture of 2nd thoracic vertebral body surrounded with paravertebral mass suggesting cold abscess or granulomatous mass at level Th1-Th3 on both sides, but with intact intervertebral disc adjacent to it. Laboratory result turned out to have normal erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) level, and increased white blood cell (WBC) count predominated with neutrophil. GeneXpert MTB assay examination from sputum of the patient producing negative result. From these examinations, patient was then diagnosed with pathological fracture 2nd thoracic vertebral body suspected due to atypical spinal tuberculosis. After that, patient was consulted to pulmonologist and started the antituberculosis drug regimen (Isoniazid, Rifampicin, Pyrazinamide, Streptomycin) 2 weeks before the surgical procedure, which is then continued for 3 months of intensive phase, and 6 months of continuation phase after the surgery.

Patient was then undergone surgical debridement, decompression - stabilization - fusion. During the exposure at level C6 through Th4, no abscess was found. Transpedicular debridement at level Th2 was then done after the insertion of pedicle screws at level C6-Th1 and Th3-Th4, and again no abscess was present, however, a granulomatous mass was present on the location of 2nd thoracic vertebral body, located on the anterior right side of the spinal cord outside the dura mater. Biopsy specimen was then extracted from the mass for histopathologic examination. The spine was then stabilized 2 dual rods and a cross-link at level Th2. Patient was then discharged after 4 days of postoperative care using Cervicothoracolumbosacral (CTLSO) orthosis.

On the 1st month after the surgery, there was improvement on both of patient's leg motor strength, which is now rated 4 on MRC scale. Patient was able to mobilize by his own, but with holding on to wall. On the 3rd month after surgery, patient has fully recovered both of his leg motor strength, which is now rated 5 on MRC scale. Patient was able mobilize on his own without any difficulties. No residual sensory disturbance is present. The pathological anatomy examination result showed granulomatous mass on the specimens from the mass and surrounding intervertebral disc with scant amount of caseating necrosis on the disc sample, sampled on the surgery. The PCR examination for the tissue sample turned out negative for tuberculosis. These confirmed the diagnosis of spinal tuberculosis on this patient due to characteristic appearance on the histopathology examination.

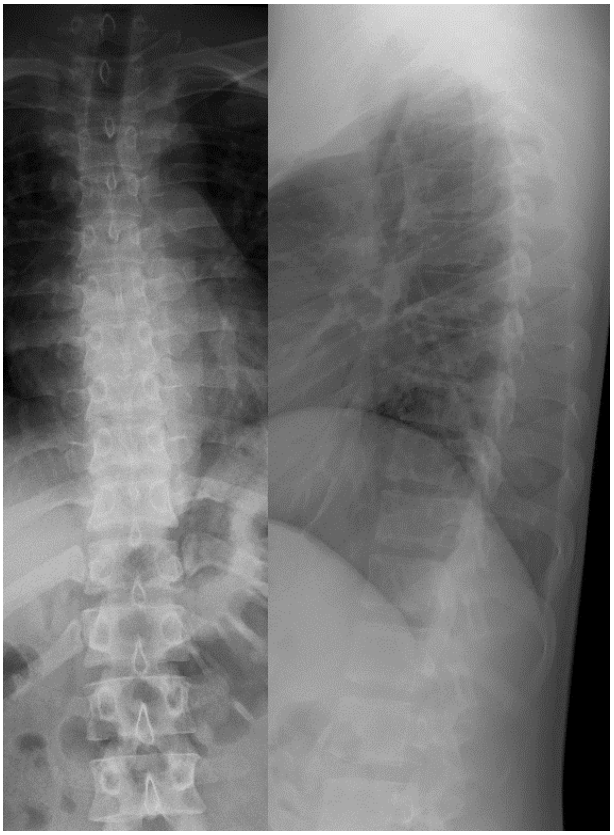

Figure 1. Preoperative X-Ray

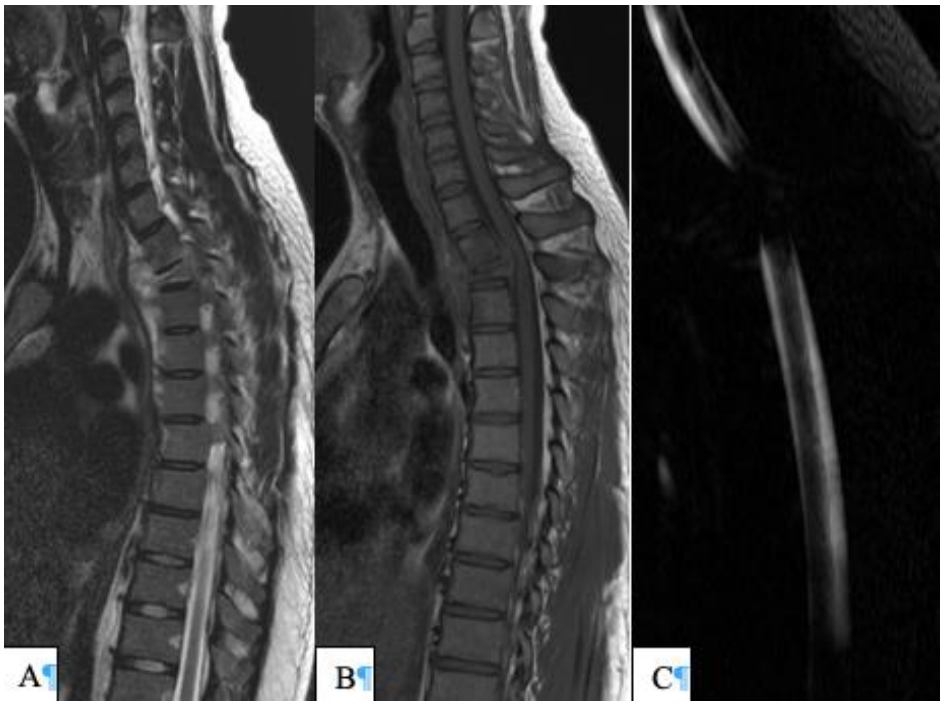

Figure 2. Sagittal View of T1W (A), T2W (B), and myelography (C)

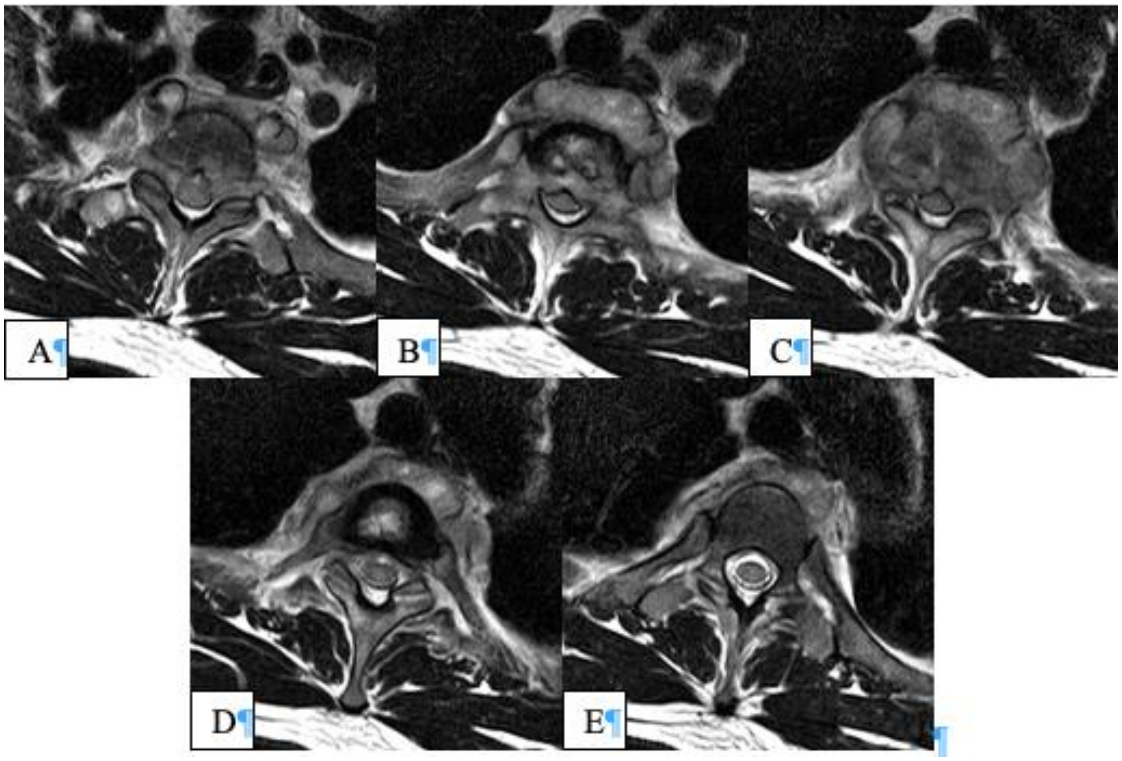

Figure 3. T2W Axial View of MRI at level of Th1 (A), Th1-2 (B), Th2 (C), Th2-3 (D), Th3 (E)

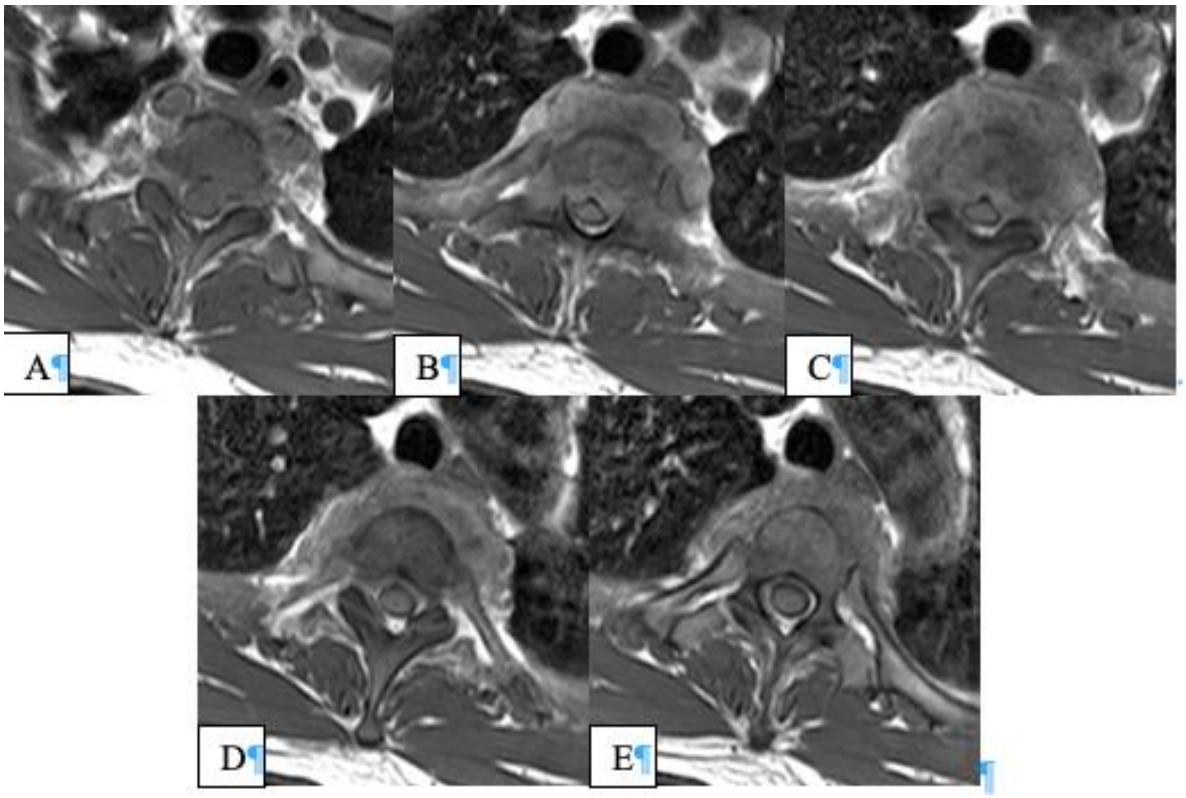

Figure 4. T1W Axial view of MRI at level of Th1 (A), Th1-2 (B), Th2 (C), Th2-3 (D), Th3 (E)

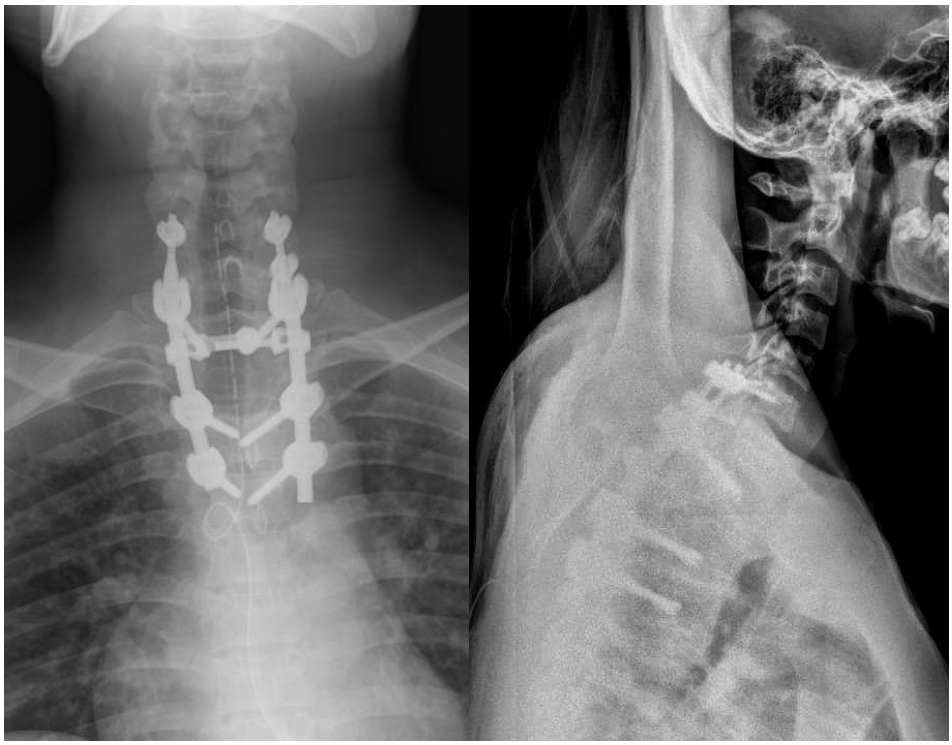

Figure 5. Postoperative X-Ray

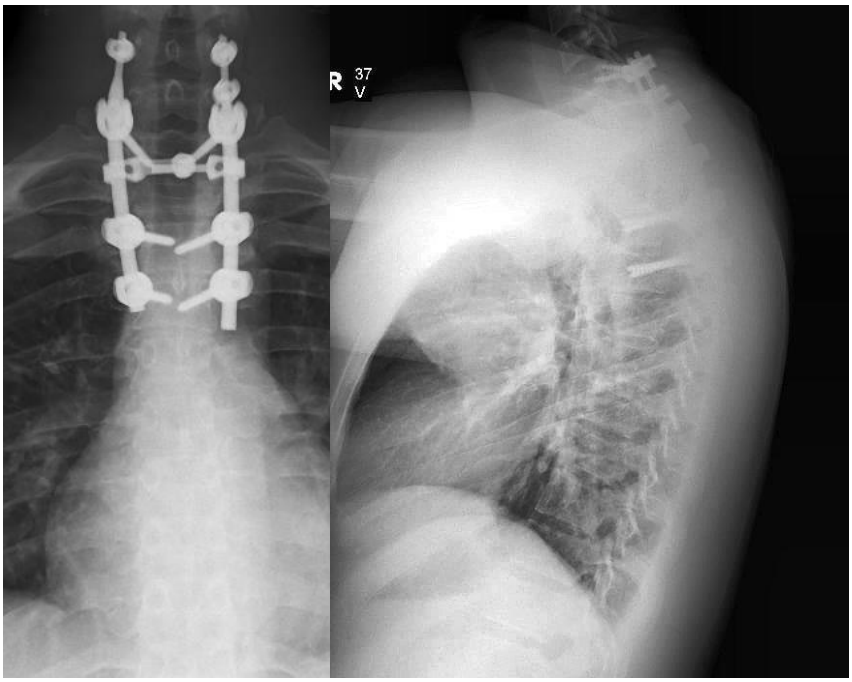

Figure 6. Control X-Ray on 3rd month after surgery

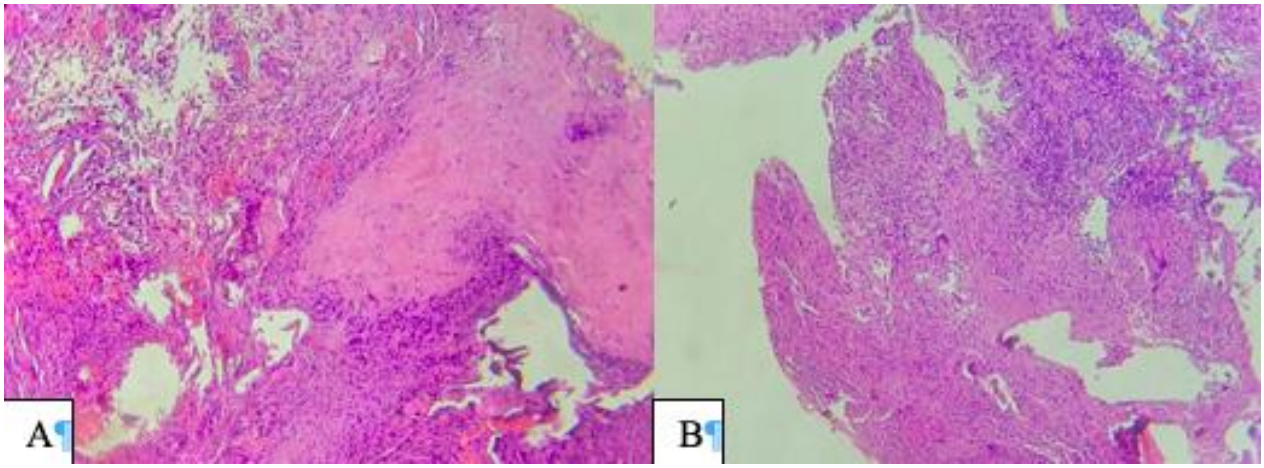

Figure 7. (A) Caseous necrosis on Lamina preparation; (B) Numerous lymphocytes, plasma cells, and Langhans giant cell

DISCUSSION

Spinal tuberculosis is the most common skeletal involvement of tuberculosis infection (Houston and Macallan, 2014). The disease starts as tuberculosis bacilli enter the vertebral body through vascular route, commonly known from Batson paravertebral venous plexus. Its valveless structure facilitate the spread of the bacilli to both lumbar and thoracal directions (Rajasekaran et al., 2018). After inoculation and multiplication on vertebral body, tuberculosis causes destruction of the bodies, initially on the endplates, extending to anterior end beneath the anterior longitudinal ligament, and later, the intervertebral disc spaces. These processes ultimately lead to anterior collapse of vertebral body resulting in deformity (gibbus) and mechanical instability accompanied with spinal

cord compression resulting in neurological deficit (Rodriguez-Takeuchi et al., 2019).

The classic spinal tuberculosis manifest as back pain, neurologic deficit, and kyphotic deformity of the spine (Khanna and Sabharwal, 2019). The patient in this case complained no back pain and no midline tenderness on the presentation. Kawsar et al. has reported one case of spinal tuberculosis without back pain, but presenting as abdominal and flank pain (Kawsar and Gopalakrishna, 2013). Similar manifestation has also reported by Meena et al., where the patient's chief complaint was lower abdominal pain without any back pain and weakness (Meena et al., 2014).

Another uncommon presentation of this patient is the upper thoracal area involvement, which is an uncommon site for spinal tuberculosis, where it is reported only in 10% of all spinal tuberculosis case (Garg and Somvanshi, 2011). This uncommon site of infection has been reported previously by Wang et al., where Th4 was involved, and was managed with debridement and stabilization using pedicle screw and 2 rods (Wang et al., 2020). This paper also reported a complication of esophageal perforation due to pedicle screw placement on 4th thoracic vertebra, which is the highest iatrogenic injury risk on the Th1-Th4 segment, where Th2 poses the highest risk (Cardoso et al., 2010)

Patients with spinal tuberculosis commonly exhibit increased ESR value and CRP concentration. A report by Kim et al., has showed that among 185 cases of spinal tuberculosis, all of them has increased ESR and CRP (Kim et al., 2019). The patient in this case has shown normal ESR (12,2 mm/hour) and CRP (3,72 mg/L). This uncommon presentation has been reported by Patel et al., where the spinal tuberculosis patient presenting, with cold abscess as its only manifestation, showed only slight increase of ESR value (28 mm/hour) (Patel et al., 2016). The research by Kim et al., also reported a favorable outcome in patients with initial lower value of ESR, but found no significant difference in outcome between higher and lower concentration of CRP on initial presentation (Kim et al., 2019). This is evident on this patient, where the patient recovered with no residual neurological deficit 3 months after the surgery.

The preoperative antituberculosis drugs was administered for 2 weeks on this patient. The efficacy of this regimen has been shown beneficial on reducing local and systemic toxins, risk of recurrence, and non-healing (Ren et al., 2016; Yang and Liu, 2013). This patient received 2 weeks of preoperative antituberculosis, and is continued for 9 months as accordance to type 1 regimen of antituberculosis drug. The good outcome this patient has may be credited to the administration of preoperative antituberculosis drug.

Absence of abscess formation coupled with granulomatous mass around the affected segment also a unique case of the spinal tuberculosis. Previously, there are several classifications of those spinal tuberculosis with atypical presentation. A paper has classified atypical spinal tuberculosis into two types: bony involvement with cold abscess, and epidural tuberculoma with no involvement of bone and cold abscess formation (Babhulkar et al., 1984). Pande et al. has further classified these atypical into two main group, those with atypical radiographic appearance, and those with atypical clinical presentation. The closest classification to this clinical presentation is the tubercular granulomas, which consists of compressive myelopathy without spinal deformity. However, this tubercular granulomas is described to have

no radiographic abnormalities, which this patient has (Pande and Babhulkar, 2002).

Negative PCR result from tissue specimen directly excised during the surgery also raise doubt on tuberculosis diagnosis. However, a review by Merino et al. has described varying level of sensitivity of PCR examination, which ranges from 38,8% up to 68,9% on confirmed spinal tuberculosis cases (Merino et al., 2012). However, this PCR technique is proven still superior to conventional visual diagnosis through staining of specimen (Portillo-Gómez et al., 2000) . This concludes that negative PCR result from tissue or pus collected during the surgery does not warrant exclusion of tuberculosis as the etiologic agent of the disease, since the golden standard diagnosis of spinal tuberculosis is still the culture and histopathology result from extracted tissue or pus specimen (Watt and Davis, 2013).

This case reported an atypical presentation of spinal tuberculosis with the sole clinical manifestation of neurological deficit below affected level. Upon further examination and management, the only confirming examination for tuberculosis is the histopathology result from specimen excised during the surgical debridement, stabilization and fusion

AUTHOR CONTRIBUTION

I Gusti Lanang Ngurah Artha Wiguna is the main author who determines the concept and review. Ryan Putra Wondany searching for literature, editing and reviews. Ida Bagus Gede Arimbawa determines the concept and review.

FINANCIAL AND SPONSORSHIP

None.

ACKNOWLEDGEMENT

None.

CONFLICT OF INTEREST

The authors declare that the study was conducted in the absence of any comercial or financial relationships that could be construed as a potential conflict of interest.

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