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CASE REPORT

EARLY DETECTION OF OSSIFICATION OF THE POSTERIOR LONGITUDINAL LIGAMENT (OPLL) IN PATIENT WITH DEGENERATIVE CERVICAL MYELOPATHY (DCM)

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Abstract

Introduction: Degenerative changes in the cervical spine leads to Degenerative Cervical Myelopathy (DCM). Ossification of posterior longitudinal ligament (OPLL) is a common cause of DCM in Asian populations and causes severe neurological deficits, which significantly impair quality of life.

Case Presentation: A 49-year-old Sunda woman presented with progressive numbness and tingling sensation followed by progressive weakness in the lower extremities. Progressive weakness then developed in the upper extremities. Physical examination revealed tetraparesis and increased reflexes. Sensory examination revealed hypoesthesia in the right T5–T12 dermatome and the bilateral L1–S2 dermatomes. She was assessed as grade 3 on the Nurick classification of myelopathy and had a Modified Japanese Orthopaedic Association (mJOA) score of 15. Non-contrast cervical magnetic resonance imaging (MRI) revealed a continuous OPLL type at C3–C5 level. She received high-dose intravenous methylprednisolone and was referred to another hospital for surgery.

Discussion: OPLL has not been documented extensively in Indonesia. Early detection of DCM can be done with an mJOA score of ≤16. Conservative treatment is recommended for minimally symptomatic patients or high-risk surgical patients. Surgical intervention has shown significant improvement in clinical outcomes, although it is not beneficial in cases of absent or minimal myelopathy (Nurick grade 1 or 2).

Conclusions: OPLL is a common cause of DCM in Asian populations. Early detection using the mJOA is paramount to achieve better recovery. Conservative treatment is used for mild DCM, while surgical intervention is used for moderate to severe DCM.

Keywords: Conservative, Degenerative Cervical Myelopathy (DCM), Modified Japanese Orthopaedic Association Score (mJOA), Nurick Classification of Myelopathy, Ossification of Posterior Longitudinal Ligament (OPLL)

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Introduction

Degenerative changes in the cervical spine lead to Degenerative Cervical Myelopathy (DCM) by causing spinal cord compression. DCM has a prevalence of 2% and can be found in 1 out of 50 adults.[1]

Ossification of the posterior longitudinal ligament (OPLL) is marked by heterotopic bone formation in the posterior longitudinal ligament and is often found as a cause of DCM, especially in Asian populations. OPLL is found primarily in individuals aged 40–60 years, in East Asian populations, and in men. OPLL leads to severe neurological deficits.[2] This significantly impairs quality of life and causes a substantial socioeconomic burden.[3] DCM caused by OPLL can have symptoms different from typical cervical myelopathy, as upper extremity symptoms are usually secondary to lower extremity symptoms.[4] This case highlights this unusual clinical manifestation, as it is important to consider in early diagnosis.

Case Presentation

A 49-year-old Sunda woman presented with progressive numbness and tingling sensations in both lower extremities, starting distally from the toes, since 1 year ago. This sensation gradually progressed, slowly rising up to the right-sided upper chest. She then gradually developed progressive weakness in the lower extremities since 10 months ago. She was initially clumsy when walking, which led to repeated falls up to 9 times while sitting. The weakness then gradually progressed, leading to weakness in both legs. She has had difficulty walking but could still walk without assistance and needs a handrail when walking up or down stairs to stabilize. She then developed progressive weakness in the upper extremities since 6 months ago. She could still button her shirt with slight difficulty and has no problems when eating with a spoon and fork. She also has had difficulty defecating since 1 year ago. She defecates every 2–3 days despite normal intake of food. She has had no problems with micturition. She had a body weight of 75 kilograms and a body height of 162 centimeters. The patient had no previous medical history. She had daily activities as a housewife. She found it difficult to do daily tasks such as sweeping, cooking, and other house chores as her symptoms progressed. She could still do house chores independently but in a limited fashion.

Physical examination revealed tetraparesis with a Medical Research Council (MRC) grade of IV in both upper extremities and an MRC grade of III in both lower extremities. Tonus examination revealed no spasticity, and atrophy was not found in any extremities. Physiologic reflexes increased in all extremities, with +3 in the biceps, triceps, brachioradial, and patellar reflexes on both sides. She also had +4 in the Achilles reflex on both sides, with clonus found during examination. Light touch examination revealed hypoesthesia in the right T5–T12 dermatome and bilateral L1–S2 dermatomes. Pinprick testing also revealed a reduced sensation in the right T5–T12 dermatome and bilateral L1–S2 dermatomes. Pathological reflexes such as Hoffman's sign and the Babinski reflex were not found during examination. She was assessed as grade 3 on the Nurick classification of myelopathy, as she had difficulty walking but could still walk without assistance. She also could still do house chores but in a limited fashion. She has a Modified Japanese Orthopaedic Association (mJOA) Score of 14, with an upper extremity motor dysfunction score

of 4, as she could still button her shirt with slight difficulty; a lower extremity motor dysfunction score of 4, as she needed a handrail to walk up or down stairs; an upper extremity sensory dysfunction score of 3, as no sensory dysfunction of the upper extremity was found during physical examination; and a sphincter dysfunction score of 3, as she had no difficulty in micturition.

Table 1. Nurick Classification of Myelopathy[5]

Grade	Description
Grade 0	No signs or symptoms of spinal cord dysfunction
Grade 1	Normal gait despite findings of spinal cord compression
Grade 2	Mild gait impairment but does not prevent employment
Grade 3	Moderate gait impairment that limits employment but does not require assistance
Grade 4	Assistance or assistive devices are required for ambulation
Grade 5	Wheelchair or bed bound

Table 2. Modified Japanese Orthopaedic Association Score (mJOA) for Cervical Myelopathy[6]

Score	Description
Motor Dysfunction	
Upper Extremities	
0	Unable to move hands
1	Unable to eat with a spoon, but still able to move hands
2	Unable to button shirt, but still able to eat with a spoon
3	Able to button shirt with great difficulty
4	Able to button shirt with slight difficulty
Lower Extremities	
0	Complete loss of motor and sensory function
1	Sensory preservation with inability to move legs
2	Able to move legs with inability to walk
3	Able to walk on flat floor with a walking aid (cane or crutch)
4	Able to walk up and/or downstairs with the aid of a handrail
5	Moderate to significant lack of stability but still able to walk up and/or downstairs without handrail
6	Mild lack of stability but still able to walk without aid with smooth reciprocation
7	No motor dysfunction
Sensory Dysfunction	
Upper Extremities	
0	Complete loss of hand sensation
1	Severe sensory loss or pain
2	Mild sensory loss
3	No sensory loss
Sphincter Dysfunction	
0	Unable to micturate voluntarily
1	Marked difficulty in micturition
2	Mild to moderate difficulty in micturition
3	Normal micturition

Table 3. Japanese Ministry of Public Health and Wellness (JPMPHW) classification system of OPLL[5]

Type	Characteristics
Localized	Single OPLL lesion with isolated vertebral level
Segmental	Multiple and separate vertebral lesions
Continuous	Single uninterrupted lesion involving multiple interspaces
Mixed	Combination of localized, segmental, and continuous

Non-contrast cervical magnetic resonance imaging (MRI) revealed OPLL at C3–C5 level, causing canal and neural foramen stenosis. MRI showed the continuous type of OPLL based on the JPMPHW classification system.[5] She received high-dose intravenous methylprednisolone and was referred to another hospital for surgery.

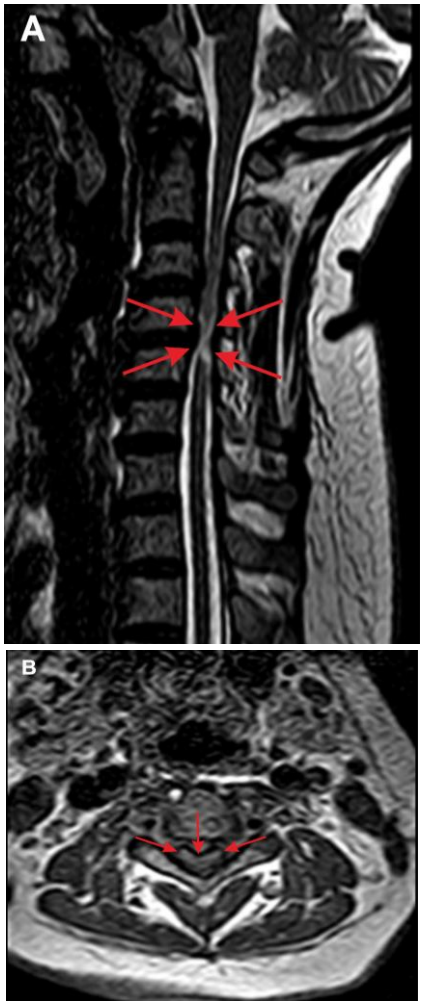

Figure 1. Non-contrast cervical MRI showing cervical myelopathy. Figure 1A shows a T2 sagittal view of the cervical spinal cord, in which hyperintensity (red arrows) can be seen, indicating cervical myelopathy. The C3–C5 spinal cord is also compressed. Figure 1B shows an axial view of the cervical spinal cord. At the C4–C5 level, hyperintensity (red arrows) can be seen mostly in the anterior region indicating cervical myelopathy

Figure 2. T1 sagittal view on non-contrast Cervical MRI showing OPLL (red arrows) compressing the spinal cord

Discussion

In OPLL, the posterior longitudinal ligament becomes thicker and less flexible, which leads to spinal compression. Approximately 70% of OPLL cases occur in the cervical spine.[2] OPLL has not been documented extensively in Indonesia or Southeast Asia. It has been found with a higher prevalence in East Asian populations (Japanese 1.9–6.3%, Korean 0.6–5.7%, Chinese 4.1%). In contrast, the prevalence of OPLL is much lower in the United States and Europe, with prevalence of 0.01–1.7%.3 Genetics play a role in OPLL pathogenesis, with involvement of genes such as RSPO2, TMEM135, WWP2, EIF3E, EMC2, PLEC, and RP11-967K21.1. Other risk factors, such as diabetes and obesity, increase the risk of OPLL formation.[7] Metabolic disorders, such as hyperglycemia, insulin resistance, dyslipidemia, calcium phosphate disturbance, and aberrant bone remodeling, are involved in the

development of OPLL.[3] This patient had a body mass index (BMI) of 28.5. She can be classified as obese class I according to the Asia-Pacific BMI classification.[8] Obesity is also a major cause of insulin resistance, which can lead to a higher risk of developing OPLL.[9] Insulin resistance has not been found in this patient because she had normal blood glucose levels; however, this process may be ongoing in this patient. This patient had an increased risk of developing OPLL because she is obese and of Sunda ethnicity. The presence of OPLL has not been extensively documented in Indonesian populations; however, a cross-sectional study showed a higher prevalence in Asian American populations, with a prevalence of 4.8% compared to Native Americans (3.2%), Africans (2.1%), Hispanics (1.9%), and Caucasians (1.3%).[3] This may indicate that OPLL has a higher risk in the general Asian population, and not just in East Asian populations. More studies are required to ascertain the risk of OPLL in the general Asian population, and this study can contribute to that cause.

Clinical manifestations of early-stage OPLL are usually cervicodynia or slightly restricted cervical range of motion. Neurological symptoms can manifest as intermittent, chronic, and progressive numbness and weakness of the extremities. Symptoms in the upper extremity are usually secondary to lower extremity symptoms, as seen in this patient.[7] This patient showed initial lower extremity symptoms with numbness, tingling, and weakness. These symptoms then gradually progressed, leading to the development of the upper extremity symptoms. This can also be seen in physical examination findings, with muscle weakness in both the upper and lower extremities. Physical examination also revealed findings of hypoesthesia and reduced sensation during the pinprick test in the right T5–T12 dermatome and the bilateral L1–S2 dermatome. The patient also had difficulty defecating, which could also be seen in OPLL.[4] These symptoms in this patient can be attributed to DCM caused by compression of the cervical spinal cord due to OPLL. These symptoms are atypical of cervical myelopathy, as symptoms usually start from the upper extremity. Some studies have analyzed the distribution of OPLL using the sum of spinal levels where OPLL is located, which is defined as the OPLL Ossification (OP) Index. A higher cervical OP index was found to have a significant association with worse lower extremity function. This signifies that the posterior column can be impaired by ossification in patients with a higher OP index. A higher cervical OP index was also associated with thoracic and lumbar OP index.[10,11] This remains a limitation in this case report, as a whole-spine MRI examination could not be done due to financial constraints. Cervical myelopathy was also associated with increased anterior pelvic tilt and lumbar lordosis, as well as decreased cervical lordosis and head flexion. This leads to less range of motion of the knee during the gait cycle, which leads to further gait disturbance and may increase lower extremity symptoms.[12] As such, it is important to consider DCM caused by OPLL in patients who develop upper extremity symptoms after initial lower extremity symptoms, especially in Asian populations.

This patient had the continuous type of OPLL based on the non-contrast cervical MRI. Different types of OPLL have been found to have different risks of developing symptoms, although there was no difference in the risk of developing myelopathy. Mixed and continuous types of OPLL are associated with a greater percentage of spinal canal stenosis compared with segmental and localized types. Neck function was found to be significantly lower in continuous types of OPLL than in mixed types. Segmental type OPLL was also more likely to be asymptomatic or have mild symptoms, such as neck pain and radiculopathy. Mixed and continuous types of OPLL were also found to have a greater risk of OPLL progression compared with the segmental type. Progression of OPLL is defined as an increase in the longitudinal or sagittal extent of the ossification by 2 mm or more, the appearance of a new lesion, and/or the bridging of 2 or more lesions to form a continuous segment.[13]

This patient has an mJOA score of 14 and thus can be categorized as having moderate myelopathy.[14] Early detection of DCM can be done with an mJOA score of ≤16, which has 90% specificity.[15] Early detection of DCM is important because earlier treatment with a conservative or surgical approach can lead to a better prognosis. As such, the mJOA score is an important tool for clinicians to help detect DCM earlier and also categorize the degree of myelopathy that has occurred to determine the best approach in treatment.

Conservative treatment with cervical traction, bracing, non–steroidal anti-inflammatory drugs (NSAID), physical therapy, and activity modification is recommended for patients with a high risk for surgery or patients with minimal symptoms.[3] Surgical intervention has shown significant improvement in clinical outcomes, although it is not beneficial in the absence of or in minimal myelopathy (Nurick grade 1 or 2).[5] Methylprednisolone was given as an adjunct to surgery because non-contrast cervical MRI and surgical intervention could not be done immediately due to the limited facility setting. Some studies have shown beneficial effects after administration of glucocorticoids in DCM.[16] This patient was stable clinically, so she did not have a high risk for surgery. She was also determined to have moderate myelopathy based on the mJOA score; therefore, conservative treatment was not done because surgical intervention was determined to be more beneficial. The patients was promptly referred to another hospital capable of surgical intervention after diagnosis

The goal of surgical intervention is spinal decompression by direct resection or thinning of the OPLL mass, or expansion of the spinal canal.[5] Surgery can be performed with an anterior, posterior, or combined approach. The anterior approach can be performed with anterior cervical corpectomy, anterior cervical discectomy, the anterior floating method, vertebral body sliding osteotomy, and anterior controllable antedisplacement. The posterior approach can be performed with laminectomy and laminoplasty. Each method has advantages and disadvantages, such as more thorough decompression and assessment, but it is accompanied by the risk of more severe

trauma, dural tear, and other complications. In this case, a posterior approach is preferred because this patient has a continuous type of OPLL. A posterior approach with laminoplasty is superior to laminectomy with fusion because it preserves cervical range of motion, maintains preoperative cervical lordosis, and minimizes postoperative neck disability.[17]

Conclusion

OPLL is a common cause of DCM in Asian populations that has not been documented extensively in Indonesia. DCM causes a significant impact on quality of life and a serious socioeconomic burden as many jobs become unavailable. Early detection using mJOA is paramount to achieve a better recovery with faster treatment. Conservative treatment is used for mild DCM, while surgical intervention is used for moderate to severe DCM.

Conflict of Interest

The authors declared no conflict of interest.

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Consent

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