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Case Report

OSSIFICATION OF THE POSTERIOR LONGITUDINAL LIGAMENT AS A PREDISPOSING FACTOR IN SPINAL CORD INJURY: A CASE REPORT

Skolastika Beatrice Tjahaja[1], Jessica Congdro[2], Andika Surya Atmadja[3]

[1]General practitioner at Gading Pluit Hospital, Jakarta
[2]General practitioner at St. Antonius Jopu Hospital Ende, East Nusa Tenggara
[3]Neurologist at Gading Pluit Hospital, Jakarta

ABSTRACT

Introduction Ossification of the posterior longitudinal ligament (OPLL) is a predisposing factor for SCI. It is discovered during imaging studies in approximately 30% SCI patients. OPLL in SCI can lead to worse neurological morbidity and result in significant reduction of quality of life if inadequately treated.

Case Summary: A 35-year-old female fell down the stairs. Despite no direct trauma to the head and neck, she complained weakness in all extremities, predominantly in both legs, accompanied with tingling and numbness. Her upper and lower motor strength were 4555/5554+ and 3111/3222, respectively. There was also hypoesthesia at and below the C5 level. Whole spine MRI demonstrated OPLL extending from C1 to T1, which impinged on thecal sac and spinal cord, especially from C3 to C7. Spinal cord edema was also identified in cervical and lumbar segments. The patient was treated with a cervical collar, intravenous methylprednisolone, and symptomatic treatments. After a week, she could stand and walk with assistance. Following 2 months of routine physiotherapy, the patient could walk and perform activities independently.

Discussion There are several factors affecting OPLL including genetic, hormonal, and environmental factors. Increase in osteogenesis-triggering growth factors is also frequently found in OPLL patients. OPLL patients have higher risk of getting SCI especially during trauma. Pharmacology treatment can be combined with physiotherapy, orthosis usage, or surgery.

Conclusion Patients with OPLL are at risk of experiencing more severe symptoms in SCI events. Appropriate therapy and management of OPLL in SCI can improve patient morbidity rates.

Keywords: SCI; OPLL; myelopathy

INTRODUCTION

Spinal cord injury (SCI) is defined as any direct or indirect trauma causing a lesion in the spinal cord, resulting in sensory, motor, and autonomic dysfunction.[1] Trauma is the leading cause of SCI, with traffic accidents accounting for 36-59% of cases, followed by falls, abuse, and other traumatic events.[2,3] According to the World Health Organization (WHO) in 2021, approximately 15.4 million people worldwide suffer from SCI.[4] SCI cases are more prevalent in males compared to females, with a ratio of 3-4:1.

Ossification of the posterior longitudinal ligament (OPLL) is the process of ossification or ectopic calcification in the posterior longitudinal ligament, most commonly occurring in cervical region vertebrae.[5] OPLL is a predisposing factor for SCI. Spinal cord injury without radiographic abnormality (SCIWORA) refers to spinal cord injury without visible vertebrae fracture or dislocation in radiography. OPLL is frequently associated with SCIWORA, as minor trauma in OPLL cases can result in SCIWORA.[6]

Although OPLL is rare, its identification is crucial. Spinal cord injury caused by OPLL can cause myelopathy, which, if left untreated may result in neurological morbidity and significant decline in quality of life.

CASE REPORT

A 35-year-old woman presented to Gading Pluit Hospital Emergency Room with paralysis in all four extremities. The paralysis suddenly occurred after she fell down a flight of stairs approximately 30 minutes prior to arrival. The patient was not paying attention to the stairs and fell in a prone position. She was unable to move her four extremities immediately afterwards. Additional symptoms included numbness and tingling sensations in her legs and arms, which were more pronounced on the left side, and generalized body pain. She denied direct injury to head or neck, loss of consciousness, headache, vomiting, and seizure. She was able to recall the entire incident.

On physical examination, the patient appeared moderately ill but was alert and oriented. Head examination revealed a 1.5x0.2 cm excoriation wound on the superior labium oris. Other examinations were unremarkable.

Neurological examination revealed a Glasgow Coma Scale (GCS) score of E4M6V5. Pupillary and cranial nerve examination were unremarkable. Motor examination showed upper extremities strength as 4555/5554+ and lower extremities strength as 3111/3222. There was decreased muscle tone in lower extremities, biceps reflex ++/++, triceps reflex ++/++, patellar reflex +/+, achilles reflex +/+. Pathologic reflex of Hoffman-Trommer -/-, babinski -/-. Sensory examination revealed hypesthesia at the Th3-4 level, more pronounced on the left side, with normal proprioception. Autonomic function was intact.

Abnormal laboratory results included hemoglobin 9.7 g/dL, hematocrit 3%, and potassium 3.2mmol/L. A non-contrast MRI of the whole spine revealed OPLL extending from C1 to T1, compressing the thecal sac and spinal cord especially at C3-C7. T2-weighted sagittal images showed spinal cord edema at the level of C3-C7 (figure 1) and L1 (figure 2).

Figure 1. Sagittal view of non-contrast cervical MRI (T2 sequence)

Figure 2. Sagittal view of non-contrast lumbar MRI (T2 sequence)

The patient was diagnosed with spinal cord injury (AIS grade C at the level of C3), cervical OPLL, anemia, and hypokalemia. Treatment included cervical collar immobilization, NaCl 0.9% 500cc, vitamin B12 5000 mcg/12 hours infusion, methylprednisolone 250 mg four times daily via IV injection, pantoprazole IV, ramosetron IV, oral pregabalin, oral paracetamol, oral rebamipide, concoction capsule (eperisone HCl, etoricoxib, amitriptilin and diazepam), and potassium chloride capsule. Vital signs and oxygen saturation were closely monitored. The patient was co-managed with internal medicine and was referred to orthopedics and medical rehabilitation. Internal medicine ordered iron deficiency anemia panel examination and results were consistent with iron deficiency anemia. Physiotherapy included general exercises and bilateral lower extremities passive range of motion (PROM) as well as passive stretching exercise 1-2 times a day.

DISCUSSION

Spinal cord injury (SCI) is defined as direct or indirect trauma resulting in spinal cord lesion which causes sensory, motor, and autonomic dysfunction. Primary injury refers to injury that occurs immediately after trauma in spinal cord. Primary injury occurs due to mechanical forces or structural compression (bone, ligaments, disc herniation, or hematoma). Secondary injury occurs due to biochemical, mechanic, and physiological changes in spinal cord which resulted from primary injury.[1] American Spinal Injury Association (ASIA) divides SCI based on the resulting degree of disability according to ASIA impairment scale (AIS)[7] (Table 1). Most common cause of SCI is trauma, especially traffic accidents, with other causes including falls, abuse, sports injuries, and surgical trauma. In addition, SCI can also be caused by malignancy or infection, with chronic tuberculosis being the most common infectious cause. Spinal cord lesion can lead to permanent disability, significant morbidity, and death.[2]

Table 1. ASIA/IMSOP SCI Classification (AIS)[1,8]

Degree	Type	Description
A	Complete	No sensory and motor function up to S4-5 segment
B	Incomplete sensory	Sensory function preserved, but motor function below lesion is affected and expanded to S4-5 segment
C	Incomplete motor	Sensory function preserved. Motor function below lesion is still preserved and more than half of the muscles below lesion has strength <3
D	Incomplete motor	Sensory function preserved. Motor function below lesion is still preserved and more than half of the muscles below lesion has strength ≥3
E	Normal	Normal sensory and motor function.

Posterior longitudinal ligament (PLL) is located along spinal canal, on the dorsal surface of vertebrae. PLL functions to prevent vertebrae hyperflexion.[9] Ossification of the posterior longitudinal ligament (OPLL) is the abnormal calcification of posterior longitudinal ligament. OPLL incident is reported to be 2.4% in Asian population and 0.16% in nonasian population. It is more prevalent in males than females. OPLL's exact mechanism is still not entirely known. Factors affecting OPLL occurrence include genetic, hormone, and environmental factors. Genetics is responsible for OPLL incidence, with several studies reporting human collagen A2 gene (COL11A2) on chromosome 6p influencing OPLL occurrence.[10,11] Increase in osteogenesis-triggering growth factor such as bone morphogenic protein (BMPs) is often found in OPLL.[9] Most commonly involved OPLL site is cervical region (70%), with symptoms frequently being myelopathy and/or radiculopathy, with severe neurologic dysfunction resulting in paralysis in extremities and immobilization, thus reducing patient's quality of life.[11] In general, cervical myelopathy symptoms progressively worsens at the age of 40-50 year old, but in 15% of OPLL cases, myelopathy symptoms can occur acutely and are often related to trauma.[12]

OPLL is usually asymptomatic, but several OPLL patients complain of continuous neck pain, numbness, radicular pain, clumsy hand syndrome and walking pattern disturbance.[13] More severe symptoms and prognosis can occur in trauma cases with more severe AIS.[6] Examination to identify OPLL includes CT scan and lateral-view X-ray. According to its lesion site, OPLL can be differentiated into 4 variants such as segmental, continuous, mixed and local (figure 3).[9] In this case report, patient was initially asymptomatic and paralysis in all four extremities only occurred after patient fell. Patient underwent cervical MRI, which showed continuous OPLL at C1 through T1, resulting in compression and edema on spinal cord especially from level C3 to C7. There was also spinal cord edema at level L1 that was assumed to be due to whiplash injury. Edema of spinal cord at L1 caused more severe paralysis in lower extremities compared to upper extremities.

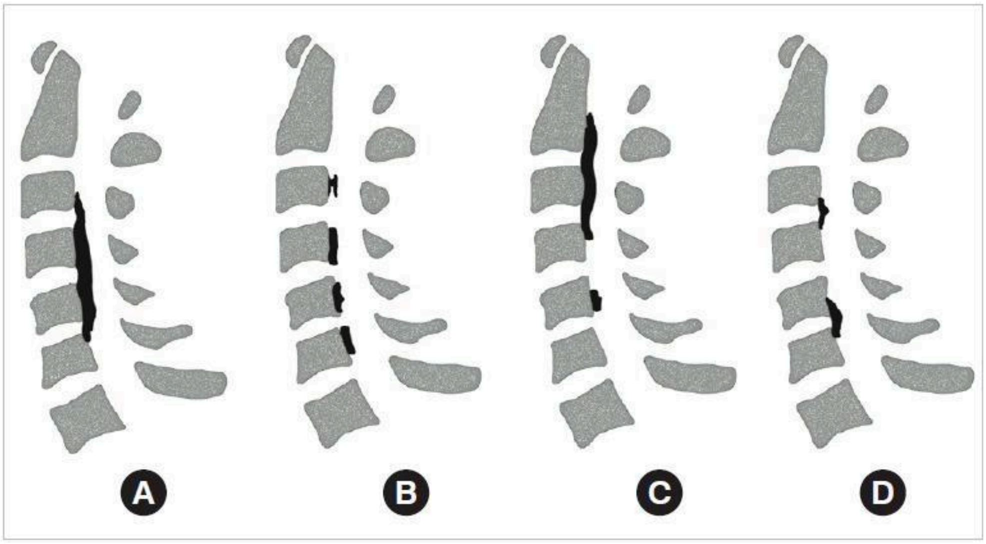

Figure 3. OPLL variation: (a) continuous; (b) segmental; (c) mixed; (d) localised.[9]

There are three main targets in treating SCI, namely maximalize neurologic recovery, spinal stabilization, and rehabilitation. SCI treatment starting with prehospital phase, can influence patients' prognosis. It starts with cervical vertebrae immobilization. Head is positioned slightly extended and with minimal distraction to prevent flexion and further spinal compression. Then, put on cervical splint or use sandbag or towel roll on lateral side or by strapping patient's head to the spine board. Airway patency was maintained, followed by shock management, while still prioritizing patient immobilization. Airway patency was kept using jaw thrust method to minimalize neck movement, nasopharyngeal airway or oropharyngeal airway can be used to maintain airway patency. Adequate ventilation is given after airway is clear. In SCI patient, it is important to note if there is respiratory failure.

OPLL treatment can be divided into two, conservative and surgical approach. Conservative treatment is chosen for mild symptomatic OPLL, by giving anti-inflammatory analgetic, muscle relaxant, vitamin B12, neuropathic pain agents, steroid, and weak opioid. In clinical practice, medical therapy can be combined with physiotherapy and orthosis. Administration of ethane-1-hydroxy-1,10 diphosphonate (EHDP) is reported to be adequately effective in preventing ossification progression by inhibiting bone calcification and resorption. Surgical treatment can be done if there are myelopathy symptoms in patient with narrow interspinal canal (<6 mm). Based on their neurological function, surgery can be done in patient with Japanese Orthopaedic Association (JOA) score of 12-13 or less. In patients with JOA 15 or higher, conservative treatment can be chosen.[14] Although OPLL is one of the predisposing factors of SCI, surgical prophylaxis in asymptomatic or mildly symptomatic OPLL patients must always consider the risks as vertebrae surgery is considered a risky procedure.

CONCLUSION

OPLL is often asymptomatic, but OPLL patients are at a higher risk of sustaining more severe symptoms in SCI following trauma even when the mechanism of injury is relatively mild. Appropriate therapy and management of SCI in OPLL can improve patient morbidity rates. Pharmacologic treatment can be combined with physiotherapy and the use of orthotic devices. Surgical intervention is considered in patients with myelopathy symptoms and narrow interspinal canal diameter.

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