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CASE REPORT

A devastating complication "white cord syndrome" following posterior cervical decompression for ossification of the posterior longitudinal ligament stenosis

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Abstract

Reperfusion injury of the spinal cord or "white cord syndrome" (WCS) refers to the sudden onset of neurological deterioration after spinal decompressive surgery. It is thought to be caused by reperfusion injury of the spinal cord. The risk of WCS might be increased in the elderly and patients with chronic hypertension as comorbidities. A 50-year-old male suggestive of a continuous ossified posterior longitudinal ligament (OPLL) with cervical spinal stenosis from C2-C4 underwent posterior decompression and posterior fixation from C2 to C4. After the patient recover from anesthesia, he developed hemiparesis on the right side with motor strength 1/5. Immediate postoperative CT scan imaging revealed no malpositioned screw, on cervical MRI disclosed a hyperintense signal in T2-weighted sequences at C3-C4 levels and the diagnosis of WCS was suspected. High dose methylprednisolone according to NASCIS II, mannitol, and methylcobalamin were given. On postoperative day (POD)-1, the patient's muscle strength improved to grade 3/5 in the upper extremity and to grade 4/5 in the lower extremity but the elbow flexion muscle strength was still 1/5. Two weeks after surgery the patient was able to walk normally but the upper extremity still no improvement. There are still no class I, II, and III evidence backing specific treatment of WCS. However, there are several prevention strategies to decrease the risk of WCS such as cerebrospinal fluid (CSF) pressure management, remote ischemic preconditioning (RIPC), intraoperative monitoring, and technique switch to laminoplasty. We recommend that the importance of early recognition and prompt treatment of white cord syndrome to minimize the complication.

Keywords: *White cord syndrome, Reperfusion injury, Ossification of the posterior longitudinal ligament, Cervical decompression*

Introduction

Ossification of the posterior longitudinal ligament (OPLL) is a structural condition of the spine in which calcification abnormally occurs at the posterior longitudinal ligament (PLL). Ossification of the posterior longitudinal ligament mostly develops in the cervical region of the

spine. In the Asian population, the incidence of OPLL is 2.4%, which is remarkably higher than in the non-Asian population which stands at 0.16%.[1] Patient with OPLL develops clinical symptoms of neurological disturbances such as pain, discomfort, numbness, and weakness of extremities from the ossification which progresses posteriorly to compress the spinal cord and nerve roots and vertically to affect the vertebral levels above and underneath.[1,2] Dynamically, the range of motion restriction caused by the ossification could also contribute to the clinical symptoms.[2]

Early cervical OPLL could be treated by conservative treatments such as the use of pain medication and anti-inflammatory drugs, but a surgical approach is recommended in the later ones, especially after myelopathy develops. The surgery could be done via anterior approach, posterior approach, or anterior-posterior approach combined. Laminectomy and fusion is one example of a posterior approach surgical method.[1,3,4]

In very rare cases, the procedure of anterior or posterior decompression could result in transient loss of motoric strength of extremities that vary in severity. The phenomenon is named "white cord syndrome", as acute reperfusion occurs in the formerly ischemic cord after decompression and leads to radiologically hyperintense findings on T2-weighted magnetic resonance imaging (MRI).[5,6] White cord syndrome is extremely rare and to our knowledge, this report is the first reported case in Indonesia.

The purpose of this report is to present a rare case of transient hemiparesis of the right side after posterior cervical decompressive laminectomy and posterior instrumented fusion at the level of C2-C4 and to review current understanding of this phenomenon. We are aiming to elucidate the community of the occurrence, pathophysiology, risk factors, prevention, and treatment of WCS.

Case Report

History and examination

A-50-year-old male presented with neck and right shoulder radiating pain for 10 years and progressive in the past year. There was no past medical history other than hypertension, which was well-controlled with medication. The family history was insignificant. He had no neurological deficit except for radiating pain of C4 and C5 dermatome. On preoperative C-spine MRI scan of the cervical spine demonstrated an OPLL mixed type from C2-C6 with an absolute canal stenosis ranging from C2 to 4. The Torg-Pavlov Ratio <0.8 and anterior posterior (AP) diameter on C2: 6.51 mm, C3: 5.37 mm and C4: 6.90 mm (Figure 1). The patient underwent an elective posterior decompressive laminectomy and posterior fixation from C2 to C4.

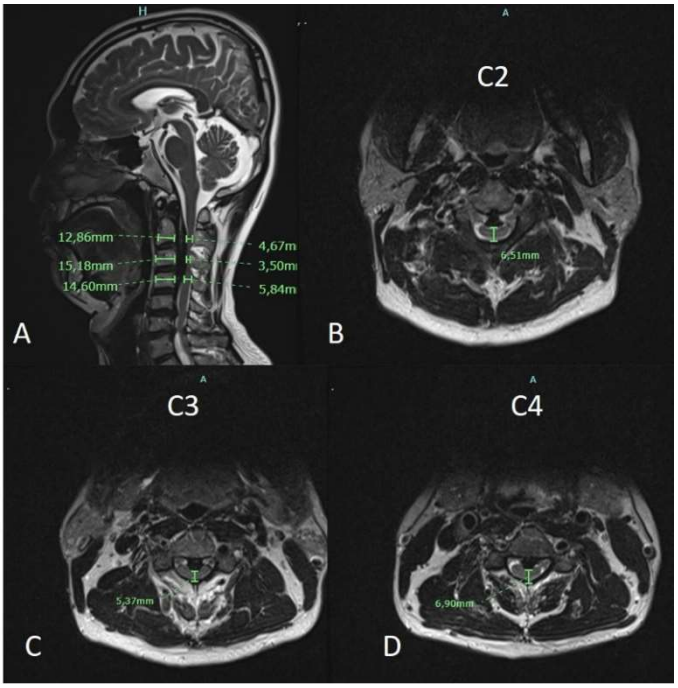

Figure 1. *(A) Preoperative sagittal T2 MRI images of the cervical spine demonstrated an OPLL mixed type from C2-6 with the Torg-Pavlov ratio on C2 to C4 < 0.8; (B) The AP diameter on C2: 6,51 mm; (C) The AP diameter on C3: 5.37 mm; (D) The AP diameter on C4: 6.90 mm.*

Operation

The patient was positioned prone with eye and abdominal pressure free and appropriate padding of all bony prominences. We didn't use intraoperative monitoring (IOM) because the IOM was used for another operation simultaneously. During the operation, the mean arterial pressure (MAP) was maintained at 85-95 mmHg. A posterior midline approach was performed with subperiosteal dissection of the affected region. Lateral mass screw fixation was done bilateral C2 to C4. A diamond high-speed burr was used at the lamina-facet junction, laminectomy was performed from C2 to C4 and the spinal cord was decompressed. The surgery was uneventful and vital signs were stable with no fluctuations, no CSF leaks or increased blood loss intraoperatively.

Postoperative course

After the operation the patient was transferred to post anesthesia care unit (PACU). The patient recovered from anesthesia and suddenly he was unable to move his right extremities with 1/5 motor strength. The patient was immediately taken to undergo computed tomography scan (CT scan) and MRI to rule out screws malposition and cord compression by hematoma. Postoperative CT scan showed

appropriately placed screws and adequate fixation of the implant (Figure 2) and no hematoma on MRI postoperatively.

However, T2-weighted MRI scans demonstrated an abnormal high signal intensity intramedullary of the spinal cord at C3 and C4 (Figure 3). White cord syndrome was highly suspected in the patient; therefore, high-dose methylprednisolone combined with mannitol and methylcobalamin were rapidly administered intravenously. The patient received high dose steroid (Methylprednisolone 30mg/kg/15min + 5.4 mg/kg/23h), according to the National Acute Spinal Cord Injury Study II (NASCIS-2) protocol. At 24-hour post treatment, the patient's muscle strength improved to grade 3/5 in the upper extremity and to grade 4/5 in the lower extremity but the elbow flexion muscle strength still 1/5. Two weeks after surgery the patient able to walk normally but the upper extremity still no improvement.

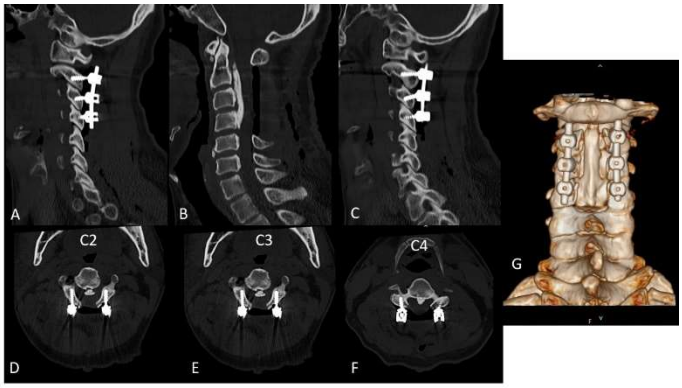

Figure 2. *(A-C) Sagittal CT scans demonstrate no malpositioned screw of C2-C4 bilaterally; (D-F) Axial CT scans of C2-C4 showed appropriately placed screws; (G) Postoperative 3D scan reconstruction.*

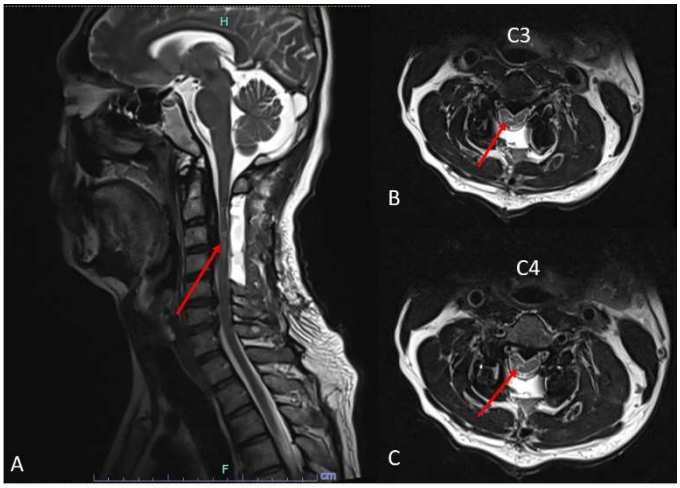

Figure 3. *Postoperative sagittal (A) and axial (B and C) T2-weighted MRI image demonstrates an abnormal intramedullary hyperintense signal at C3 and C4 (Red Arrow). The appearance of WCS.*

Discussion

White cord syndrome is an extremely rare case of reperfusion syndrome which occurs at the chronically ischemic sites of the spinal cord after decompression without any remarkable intraoperative events. The phenomenon manifests clinically as acute transient neurological deterioration in muscle strength, causing paresis or plegia in few or all extremities.[6,7] The epidemiology of WCS is not thoroughly clear as it is very few in number of cases and much fewer in number of reported ones. Until the day this review was written, there are no more than 20 reported cases of WCS as can be seen as Table 1.

The initial case of WCS was reported in 2013 by Chin *et al.,* when a 59-year-old male developed tetraplegia after undergoing anterior cervical discectomy and fusion (ACDF) without any significant intraoperative events. Post-operative MRI of the patient showed large area of hyperintensity changes of the affected spinal cord on T2-weighted MRI after 3 days, which first brought out the name “white cord syndrome”. The patient was monitored for months and had successfully reached American Spinal Injury Association (ASIA) score D after 16 months, significantly better than the ASIA score B he had immediately after surgery.[6,8] The second reported case is published 4 years later by Giammalva *et al.* when a 64-year-old male developed severe tetraparesis with complete paraplegia after undergoing the similar surgery as the patient of Chin *et al.,* Similar findings and progression of clinical symptoms are also found after post-operative follow-up.[8,9] In 2018, Antwi *et al.,* published the first case of WCS that occurred after surgery of posterior approach. The patient is a 68-year-old male developed hemiparesis of the left side after posterior cervical decompressive laminectomy and fusion following diagnosis of cervical spondylotic myelopathy.[10] In all of previously reported cases, the acute post-operative neurologic deterioration mostly improved progressively after pharmacotherapy of high-dose intravenous (IV) steroid and physiotherapy.[5–16]

The pathophysiology of WCS is still not completely understood. Various hypotheses are being studied around the fact that the decompression of the spinal cord is achieved, which means that there is no longer any significant structural stress. The absence of the direct insult signifies vascular contribution, resulting in the hypothesis of involvement of microthrombi, occlusion of small artery or anterior spinal artery, altered perfusion caused by internal recoil of spinal structure, and reperfusion injury caused by ischemia.[17] The latter is currently the most studied etiology of WCS with the vastest reports and publications.[5–16]

Table 1. *Demographic and Clinical Characteristics of previous reported cases of WCS5–16*

Author (Year)	Case Amount	Age	Sex	Preoperative Symptoms	Procedure	Post-Operative Symptoms	Treatment	Outcome
Chin *et al.,* (2013)[8]	1	59	Male	Severe cord compression, neck pain, radiculomyelopathy, ataxia	ACDF C4-C5, C5-C6 and plating	C6 incomplete tetraplegia	Extensive decompression and C5 corpectomy, high-dose IV hydrocortisone, and physiotherapy	Bilateral upper limb strength improved to full strength except for 3+/5 left finger flexion, extension, and interossei. Right lower extremity improved to full strength. Left lower extremity 3/5
Giammalva *et al.,* (2017)[9]	1	64	Male	Severe neck pain irradiated to both arms, gait disturbance, urinary incontinence, tetraparesis	ACDF C3-C4, C5-C6 and interbody cage	Worsening of tetraparesis	High-dose IV dexamethasone and physiotherapy	Right hand extension 3/5 Right arm flexion 2/5 Both leg flexion 2/5
Antwi *et al.,* (2018)[10]	1	68	Male	Intermittent paresthesias, numbness, intermittent balance difficulties, and pain in the bilateral upper extremities.	Posterior C4-C7 laminectomy and lateral mass fixation at C3-C7	Left hemiparesis	C7 laminectomy and screw removal, high dose IV solumedrol followed by dexamethasone and physiotherapy	Left wrist flexion 3/5 Left wrist extension 4+/5 Left elbow flexion 4+/5 Left elbow extension 4+/5 Left hip flexion 2+/5 Left knee extension 4/5 Left ankle dorsiflexion 1/5 Left ankle plantar flexion 2/5
Vinodh *et al.,* (2018)[11]	1	51	Female	Progressive paraparesis and acute urinary retention	Posterior C2-C5 laminectomy 1and pedicle screw fixation at C1-C2, C5-C6	Tetraplegia, C3 sensory loss	High-dose IV dexamethasone and physiotherapy	No improvement
Papaioannou *et al.,* (2019)[12]	1	79	Male	Gait disturbance, neck pain, hemiparesis	Posterior C3-C6 laminectomy and lateral mass fixation at C2-C7	C6 incomplete paraplegia	Extensive decompression, high-dose IV steroid, and physiotherapy	Right upper extremity 3/5 Left upper extremity 4/5 Right leg 2/5 Left leg 3/5
Wiginton *et al.,* (2019)[5]	1	41	Male	Low back pain, burning sensation in bilateral lower extremities, weakness in bilateral upper extremities	Posterior C1 laminectomy	Tetraplegia	High-dose IV dexamethasone and physiotherapy	Complete resolution
Mathkour *et al.,* (2020)[13]	1	79	Male	Bilateral lower extremity weakness, difficulty using hands	Posterior C3-C5 laminectomy and lateral mass fixation at C2-C6	Worsening of tetraparesis	High-dose IV dexamethasone and physiotherapy	Full strength except right biceps 4/5, right triceps 4/5, right hand 3/5, right wrist 2/5, and right interossei 2/5
Jun *et al.,* (2020)[14]	1	49	Female	Neck pain, second and third finger radiating pain	ACDF C6-C7	Paraplegia	C4-C7 laminoplasty, high-dose IV methylprednisolone , and physiotherapy	Both lower limb full strength
Liao *et al.,* (2020)[7]	1	51	Male	Numbness of bilateral upper extremities, unsteady gait	Posterior C3-C4 laminectomy and pedicle screw fixation at C2-C5	Left hemiparesis, neck pain and swelling	High-dose IV methylprednisolone , hyperbaric oxygen therapy, and physiotherapy	Complete resolution
Busack *et al.,* (2020)[15]	1	63	Male	Progressive difficulty walking, declining balance, neuropathic pain in extremities	Posterior C3-C6 laminectomy and fusion at C2-T1	Tetraplegia, T3 sensory loss	High-dose IV dexamethasone and physiotherapy	Full strength in all extremities except bilateral dorsiflexion 4/5 T10 sensory loss
Fathalla *et al.,* (2020)[6]	7	61-70	5 males and 2 females	Nurick scale of 1-3	Posterior laminectomy at various levels between C3-C7 and variation in fusion/no fusion	Neurological deterioration	IV steroid and physiotherapy	Nurick scale of 3-4
Singh *et al.,* (2021)[16]	2	59 and 66	2 females	Patient 1: Progressive neck pain, tingling sensation in both legs, spastic gait Patient 2: Walking disturbances, tingling sensations in both hands, deterioration of fine motor skill	Posterior C3-C6 laminectomy and anterior C4 partial corpectomy, C6-C7 discectomy	Patient 1: Tetraparesis Patient 2: Tetraparesis and urinary incontinence	High-dose IV dexamethasone and physiotherapy	Patient 1: 4/5 in all extremities Patient 2: Left arm more than 3/5 and normal micturition

The mechanism of ischemic-reperfusion injury begins with the decompression procedure that releases the formerly chronically compressed spinal cord. Expansion of the spinal cord then takes place to allow the ischemic tissues to receive their required blood. The re-expansion induces trauma along the blood-spine barrier. The trauma could be caused directly by the flow of the blood or indirectly by the hazardous contents of the blood, such as oxygen free radicals and lipid peroxidation of the neuronal membrane.[18] Observation in rats concluded a significant increase of blood flow to the spinal cord after decompression procedure, with better clinical outcomes in rats with early decompression than delayed one. Another additional experiment using 8-oxoG DNA to see directly proportional increase in reactive oxygen species showed higher number of 8-oxoG DNA in rats that underwent spinal decompression compared to the others that still had spinal cord compression. Oxygen free radicals disturb axons of the spinal cord via oxidative stress which results in increased inflammatory markers such as tumor necrosis factor α (TNF-α) and interleukin-1 β11. Lipid peroxidation of the neuronal membrane could also cause a further secondary injury-induced degenerative cascade.[5]

The risk of WCS might be increased in elderly and patients with chronic hypertension as comorbidities. The average of age in patients from our literatures is 62 years old with only 2 cases found under the age of 50. We also found 9 cases of hypertension in previous cases. Our patient also had hypertension which is controlled by daily β-blocker medication. Old age appears to be involved with impaired immune system which leads to increased chronic oxidative stress, while hypertension directly disrupts blood-spine barrier by causing atherosclerosis and endothelial damage which increases permeability to inflammatory mediators.[6]

There are several prevention strategies to decrease the risk of WCS. They are CSF pressure management, remote ischemic preconditioning (RIPC), intraoperative monitoring, and technique switch to laminoplasty.[7] Spinal cord perfusion is dependent on the flow of CSF. Therefore, controlling the CSF pressure could be used to manage perfusion of the spine. CSF drain could be placed pre-operatively to reduce the pressure of severely compressed spinal cord. Hypothetically, gradual reduction of CSF pressure before decompression procedure might help inhibit abrupt reperfusion and edema which happens immediately after.[15] Remote ischemic preconditioning is a technique to induce ischemic tolerance in specific tissues, distant tissues, and even distant organs by creating 3 cycles of right upper arm ischemia, achieved by inflating a cuff on right upper arm to 200 mmHg for 5 minutes and repeating it 3 times with two 5-minute intervals. The mechanism of RIPC is associated with the reduction of spinal injury biomarkers such as neuro-specific S-100B and enolase.[19] Intraoperative neurophysiological monitoring should be used to evaluate spinal cord injury via somatosensory evoked potential (SSEP) and motoric evoked potential (MEP) alteration. Laminoplasty is also hypothesized to avoid excessive spinal cord expansion compared to laminectomy which is commonly used before the incidence of WCS in previously reported cases.[7]

There are still no class I, II, and III evidence backing specific treatment of WCS. However, most of the patients in previously reported cases clinically improved after the treatment of high-dose intravenous corticosteroids for approximately 48 hours, MAP maintenance of more than 85 mmHg for 5-7 days, and early physiotherapy. Corticosteroids such as methylprednisolone and dexamethasone are chosen as the main pharmacotherapy for WCS to inhibit inflammation and lipid peroxidation following the most likely etiology of ischemic-reperfusion injury.[5,11,16] Reduction of MAP in WCS is contributed to reduction in spinal cord edema, considering the increase of complication and worse outcomes in hypertension after mechanical thrombectomy for large vessel occlusion which has a technically similar open reperfusion phenomenon. Although the minimum MAP of 85 mmHg is used in many cases, the maximum target of MAP in the treatment of WCS is still undetermined.[15] Rehabilitative physical therapies are given in accordance with the severity of the motoric weakness and extremities involved. Passive range of motion (ROM) exercises are applied to plegic patients with additional active ROM-and-strengthening exercises for patients with paresis. Range of motion exercises help in preventing complications such as contracture and stiffness, whereas muscle-strengthening exercises help in maintaining and progressively improving motoric strength.[20]

Conclusion

Surgeons should explain the possibility of white cord syndrome before cervical decompression surgery, intraoperative monitoring is highly recommended, and surgeon should perform a neurological examination immediately after the patient wake-up. We recommend that the importance of early recognition and prompt treatment of white cord syndrome to minimize the complication.

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The author declare that they have no conflict of interest.

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