Case Report

Misdiagnosis of “White Cord Syndrome” following posterior cervical surgery for ossification of the posterior longitudinal ligament: A case report

Shankar Acharya, Deepak Kaucha, Arashpreet Singh Sandhu, Nitin Adsul, R. S. Chahal, K. L. Kalra

Department of Ortho-Spine Surgery, Sir Ganga Ram Hospital, New Delhi, India.

E-mail: Shankar Acharya - spineshankar@gmail.com; Deepak Kaucha - i201421337@gmail.com; Arashpreet Singh Sandhu - arash.sandhu@gmail.com; *Nitin Adsul - no1.nitinadsul@gmail.com; R. S. Chahal - rupinder72@hotmail.com; K. L. Kalra - kashkalra55@gmail.com

*Corresponding author:
Nitin Adsul,
Department of Ortho-Spine Surgery, Sir Ganga Ram Hospital, New Delhi, India.
no1.nitinadsul@gmail.com

ABSTRACT

Background: Following decompressive cervical surgery for significant spinal cord compression/myelopathy, patients may rarely develop the “White Cord Syndrome (WCS).” This acute postoperative reperfusion injury is characterized on T2W MRI images by an increased intramedullary cord signal. However, it is a diagnosis of exclusion, and WCS can only be invoked once all other etiologies for cord injury have been ruled out.

Case Description: A 49-year-old male, 3 days following a C3-C7 cervical laminectomy and C2-T1 fusion for extensive cord compression due to ossification of the posterior longitudinal ligament (OPLL), developed acute quadriparesis. This new deficit should have been attributed to an intraoperative iatrogenic cord injury, not the WCS.

Conclusion: Very rarely patients sustain postoperative significant/severe new neurological deficits attributable to the WCS. Notably, the WCS is a diagnosis of exclusion, and all other etiologies (i.e. intraoperative iatrogenic surgeon-based mechanical cord injury, graft/instrumentation extrusion, failure to adequately remove/resect OPLL thus stretching cord over residual disease, other reasons for continued cord compression, including the need for secondary surgery, etc.) of cord injury must first be ruled out.

Keywords: Cervical decompressive surgery, Cervical myelopathy, Ossification of the posterior longitudinal ligament, White cord syndrome

INTRODUCTION

The cervical “White Cord Syndrome (WCS)” is attributed to an acute intraoperative “reperfusion injury.” This is characterized on the postoperative T2W MRI by an increased intramedullary cord signal.[9] However, WCS is a diagnosis of exclusion, and all other “aetiologies” must first be ruled out (i.e. no residual cord compression, no disruption/intrusion/extrusion of a graft/plate/instrumentation, no intraoperative mechanical-dissection related cord injury sustained secondary to poor technique, no residual ossification of the posterior longitudinal ligament (OPLL)/disc/other pathology, and others potential “traumatic factors which may or may not require additional surgery”).[2,4,5] Here, we present a patient with cervical OPLL who, following a cervical laminectomy/fusion from C2-T1 developed a 3-day delay quadriparesis. This new deficit was first mistakenly attributed to the WCS, rather than to an intraoperative iatrogenic cord injury.
**CASE REPORT**

A 49-year-old male presented with 8 years of progressive myelopathy; he had acutely deteriorated over the prior month. On examination, he exhibited a significant left hemiparesis accompanied by marked hyperreflexia (i.e. left-sided Hoffman's and Babinski response). The cervical CT and MR scan both documented OPLL extending from C2-C3 to C7-T1 resulting in significant cervical canal stenosis/cord compression [Figure 1a]. He underwent a C3-C7 laminectomy with C2-T1 lateral mass/ pedicle screw fusion. Unfortunately, this was performed without intraoperative neural monitoring (IONM) (i.e. no somatosensory evoked potentials, motor evoked potentials, or electromyography). Without IONM, the patient likely sustained a traumatic but unrecognized intraoperative iatrogenic cord injury.

**Postoperative course**

On postoperative day 1, his strength was 4/5 in the upper, and 5/5 in the lower extremities, and his spasticity had decreased. However, on postoperative day 3, he became acutely quadriparietic (i.e. 2/5 in the left and 3/5 in the right upper extremity, with 4/5 motor function in both lower extremities without an accompanying sensory deficit).

**Postoperative imaging**

The postoperative MRI documented adequate cord decompression, but new intramedullary cord edema on the T2-weighted image opposite the C3, C6, and C7 levels [Figure 1b]. Notably, these abnormal high intrinsic cord signals were all located directly opposite foci of maximal preoperative OPLL-related cord compression. Therefore, the patient most likely sustained an iatrogenic traumatic cord injury and the deficit was not attributable to the WCS.

**Postoperative management**

Once the deficit appeared, the patient was started on high-dose intravenous steroids (Methylprednisolone). No further surgery was performed (i.e. no surgical lesion was identified on the postoperative MR). The patient regained his preoperative neurological baseline within 7 postoperative days.

**DISCUSSION**

The WCS is rare following cervical spine surgery. It is characterized by an increased intramedullary cord signal on postoperative T2W MRI scan. The pathophysiological

### Table 1: WCS following posterior cervical surgery.

| Author          | Primary pathology                      | Postoperative symptoms                  | Steroids | Outcome                  |
|-----------------|----------------------------------------|------------------------------------------|----------|--------------------------|
| Vinodh et al. (2018) | C3 metastatic tumor extra/intradural extension | Quadriplegia | Yes      | No improvement (6 weeks) |
| Antwi et al. (2018)  | C4-6 spondylosis stenosis C4-5 listhesis | Loss of sensation below C3 Paraplegia (0/5 on LE) | Yes      | Partial recovery         |
| Papaoannou (2019)  | C4-C6 stenosis                         | Paraparesis                              | Yes      | Persistent weakness (18 months) |
| Wiginnton et al. (2019) | C1 stenosis                          | Quadriplegia                              | Yes      | Not mention              |
| Mathkour et al. (2019) | C3-6 spondylosis with canal stenosis | Right hemiparesis                        | Yes      | Partial recovery (4 months) |
| Liao et al. (2020) | C2-4 OPLL                               | Left hemiparesis                         | Yes      | Recovery to Preoperative level Re-exploration at 2 months for continued CSF leakage |
| Our case         | C2-C3 to C7-T1 OPLL                    | Quadriparies                             | Yes      | Recovered (8 weeks)      |

OPLL: Ossification of the posterior longitudinal ligament, WCS: White cord syndrome
mechanism of WCS is a reperfusion injury; it occurs due to the sudden expansion of a previously markedly compressed cord. Following an acute decompression, the chronically ischemic cord becomes suddenly exposed to an acutely increased blood supply. This leads to disruption of the blood-spinal cord barrier and triggers an inflammatory cascade releasing free radicals, and promoting cord/neuronal damage.[4,5]

WCS diagnosis of exclusion

The diagnosis of WCS is one of exclusion; all other potential aetiologies of injury must first be ruled out.[2,3,7,8] In 2004, Seichi et al. diagnosed WCS in 7 out of 114 patients following decompressive surgery; four had worsening of symptoms/signs that never recovered, while three improved despite the expansion of an increased intrinsic cord signal on the postoperative MRI.[7] Vinodh et al. in 2018 diagnosed the WCS in a patient with a C3 vertebral body metastatic tumor who postoperatively worsened, and failed to respond to high-dose steroids.[8] Several other authors similarly reported WCS following cervical surgery involving acute decompressions (i.e. laminectomy/fusions), while only one case involved a patient with OPLL.[1,4-6,8,9] In the case presented, the patient's delayed deficit following OPLL surgery was most likely attributable to an iatrogenic intraoperative cord injury rather than the WCS. Notably, the WCS should only be invoked once all other causes for postoperative neurological deterioration have been identified. Further, the opportunity to "reverse" any treatable findings should not be missed.

CONCLUSION

The WCS is a diagnosis of exclusion that only rarely follows cervical spine surgery. However, it is a diagnosis of exclusion, and all other causes of intraoperative and postoperative cord injury must first be ruled out.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

Financial support and sponsorship

Nil.