Clinical Case Report

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# A rare, acute neurologic deterioration associated with the overactive autoimmune response of ankylosing spondylitis after cervical laminoplasty

## A case report

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**Abstract**

**Rationale:** We report a rare, acute neurological deterioration after cervical laminoplasty due to post-decompression spinal cord edema associated with ankylosing spondylitis in a 52-year old male patient. The patient was diagnosed with cervical spondylotic myelopathy due to ossification of the posterior longitudinal ligament which was complicated by ankylosing spondylitis. A cervical laminoplasty was performed, adversely resulting in paraparesis and loss of tactile sense. An emergency CT scan following the first laminoplasty revealed that the spinal cord compression due to spinal cord swelling and limited-expansion in cervical canal space. The abnormal pathological state of ankylosing spondylitis may have aggravated spinal cord re-perfusion and increased edema after decompression.

**Patient concerns:** Paraparesis and loss of tactile sense after the surgery immediately.

**Diagnoses:** Acute neurological deterioration after cervical laminoplasty.

**Interventions:** A second emergency surgery was performed to remove the C2-C5 laminae.

**Outcomes:** Six months later, the patient had experienced slight improvement in neurological function.

**Lessons:** Abnormal spinal cord immune inflammatory reaction associated with ankylosing spondylitis and limited decompression may lead to acute neurological deterioration. The potential overactive inflammatory response following surgery in the patients with autoimmune rheumatoid disease should be carefully considered in spinal surgery. Timely diagnosis and treatment may benefit these patients.

**Abbreviations:** AS = ankylosing spondylitis, CRP = C-reactive protein, ESR = erythrocyte sedimentation rate, OPLL = ossification of the posterior longitudinal ligament.

**Keywords:** acute neurologic deterioration, ankylosing spondylitis, cervical laminoplasty, overactive autoimmune reaction

## 1. Introduction

Acute neurologic deterioration after spine surgery is a serious complication which may lead to surgical failure and may be devastating for patients and their families.[1–4] Fortunately, this complication is rarely seen in the clinic, especially in posterior cervical surgery. Cervical laminoplasty is a safe and effective posterior decompression procedure for multilevel cervical canal stenosis.[5,6] Though the neurologic deterioration is typically second to epidural hematoma and cervical canal restenosis or hinge collapse has been reported during long-term follow-up,[3,7] there are a few reports of acute neurologic deterioration associated with rheumatic disease in cervical laminoplasty. Here we report an acute neurologic deterioration after cervical laminoplasty in a patient with ankylosing spondylitis (AS).

## 2. Case report

This study was approved by the medical ethical committee of our hospital. The patient signed the informed consent for the publication of his clinical and radiologic data.

### 2.1. Preoperative information

A 52-year-old man walked to our hospital to search for help because of "Extremities weakness for 7 years, progressing to an unstable gait for 4+ months." The medical history included lower back pain over 20 years and could gain relieved after exercise. The physical examination revealed obvious limitation of neck movement, and the impairment of superficial sensation in extremities was also documented. The muscle forces to flex left elbow, extensor left elbow, dorsiflex left wrist, and abduct left little finger were grade 3 according to muscle strength grading system.[8] The muscle forces to flex right elbow, extensor right elbow, and dorsiflex right wrist were grade 4. The muscle strengths of lower limb muscles were grade 5. The preoperative plain film standing X-ray of the whole spine and pelvis showed typical bamboo spine changes (Fig. 1). HLA-B27 was negative. The erythrocyte sedimentation rate (ESR) and C-reactive protein

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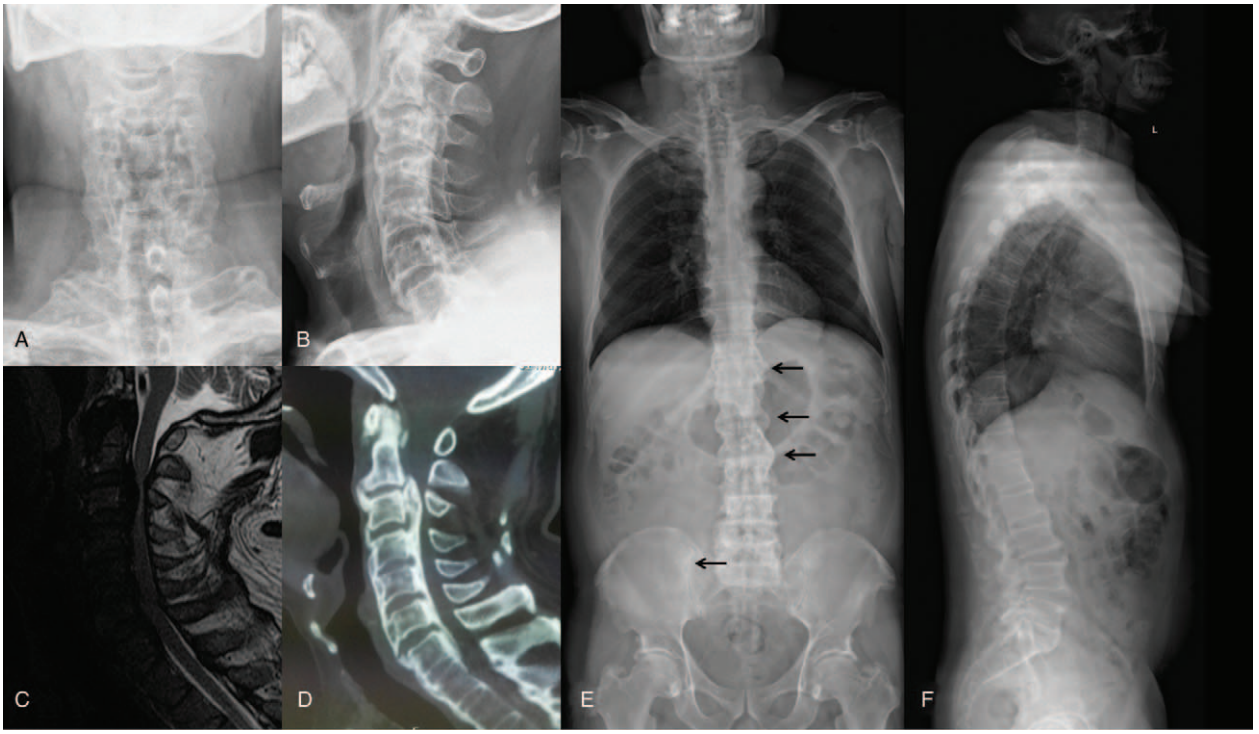


**Figure 1.** Preoperative radiographic images. (A) Cervical neutral X-ray radiographs. (B) Cervical lateral X-ray radiographs. (C) Midsagittal magnetic resonance image. (D) Midsagittal 3-dimensional reconstruction computed tomography image. (E) Standing full-spine neutral X-ray radiographs. ←, the typical sacroiliitis and bamboo spine changes. (F) Standing full-spine lateral X-ray radiographs.

(CRP) were normal. Cervical spine magnetic resonance imaging (MRI) showed continuous ossification of the posterior longitudinal ligament (OPLL) with cervical spinal canal stenosis from C3 to C7. The Pavlov values from C3 to C7 were 0.19, 0.46, 0.52, 0.49, and 0.54, respectively. The occupy ratios of OPLL from C3 to C7 were 74%, 46%, 41%, 34%, and 31%, respectively. Other medical comorbidity included hypertension, fatty liver, and renal cysts. The main clinical diagnoses were cervical spondylotic myelopathy, OPLL, and AS.

### 2.2. First surgery

The patient underwent a C3-C7 laminoplasty with mini-plate fixation companied with C2 dome semilaminectomy under general endotracheal anesthesia. The procedures were successful, and the spinal cord evoked potential monitor showed no change during the surgery. However, following recovery from the anesthesia, the patient complained of paraparesis and loss of tactile sense. Physical examination revealed that the muscle strengths of lower limb muscles were grade 0, and the muscle strengths of upper limbs had decreased to grades 1 and 2. An emergency computed tomography (CT) scan documented substantial swelling of the spinal cord, and spinal cord compression at the level of C2/C3 (Fig. 2).

### 2.3. Second surgery

An emergency surgery was immediately scheduled. A posterior surgery along the original approach was performed. During the second surgery, the cervical spinal cord was observed to be markedly swollen and the spinal dura mater was contacting the C2-C5 laminae. After removing the mini-plate on C3-C5, the dura mater bulged out of the cervical spine canal. The laminae of C2-C5 were then removed. Intraoperative exploration found the spinal cord was distended, with no compression at the level of C6 and C7. Cerebrospinal fluid leakage was observed following surgery. Postoperative treatments included methylprednisolone to reduce inflammation and spinal cord edema, anti-infection therapy, neuro nutrition treatment, and effective drainage to reduce the cerebrospinal fluid. Four days after the second surgery, the muscle strength in the upper limbs recovered back to grade 3 and the muscle strength in lower limbs recovered back to grade 4. Eleven days after the second surgery, the drainage tube was removed, and the wound healed.

### 2.4. Follow-up information

At the 6-month follow-up, the patient had a slight improvement in neurologic function compared to before the surgeries. The Japanese Orthopaedic Association scores improved from 10 before the surgeries to 12 at the follow-up. The patient had no complaints of discomfort such as axial pain or C5 palsy.

## 3. Discussion

Posterior cervical laminoplasty is recommended as a safe and effective procedure to treat multilevel cervical spine canal stenosis.[6] Few studies have reported postoperative acute neurologic deterioration. Ahn and Fehlings[1] reported a 2.2% incidence of neurologic injury during posterior cervical surgery, which commonly presented as nerve root or spinal cord injury caused by posterior cervical instrumentation, such as lateral mass

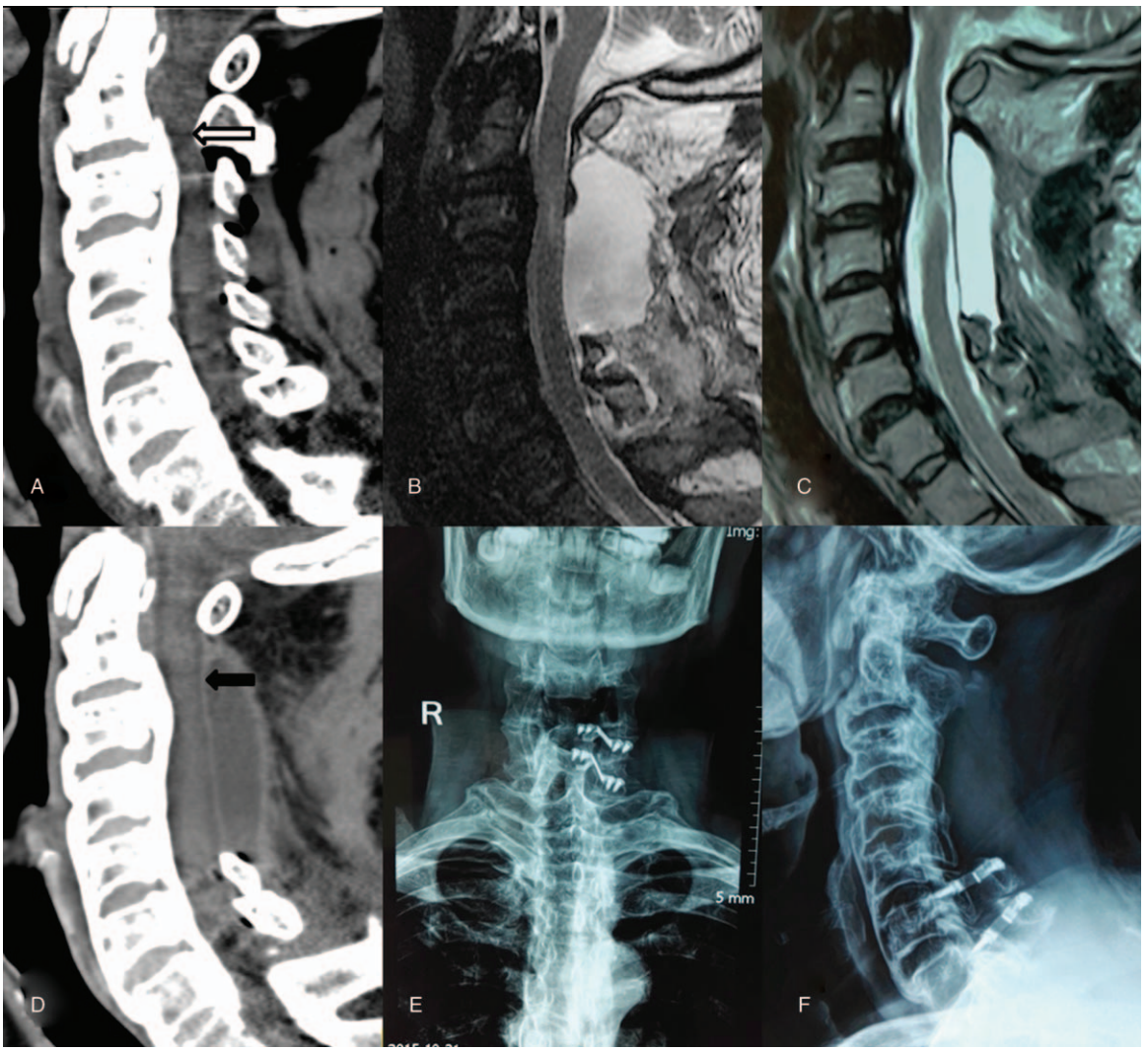


**Figure 2.** Postoperative radiographic images. (A) Midsagittal 3-dimensional (3D) reconstruction computed tomography (CT) image after first surgery. The spinal cord was edema and was compressed at C2/C3 level (⇦). (B) Midsagittal magnetic resonance image after second surgery immediately. The compression was removed but the spinal cord was still swollen. An epidural cyst formed because of cerebrospinal fluid leakage. (C) Midsagittal magnetic resonance image at 6-month follow-up. The spinal cord edema disappeared. The epidural cyst still existed. (D) Midsagittal 3D reconstruction CT image at 6-month follow-up. The spinal cord at C3/C4 level was not compressed (⬅). (E) Cervical neutral X-ray radiographs at 6-month follow-up. (F) Cervical lateral X-ray radiographs at 6-month follow-up.

screws. Seichi et al[3] reported that 18 out of 237 patients with OPLL suffered deterioration of lower-extremity function after laminoplasty across 27 individual institutes. The overall rate of neurologic deterioration was 3.1%. In their study, the causes of deterioration included epidural hematoma, spinal cord herniation through injured dura mater, and incomplete laminoplasty due to vertebral artery injury while making a trough. The potential mechanism of acute neurologic deterioration after cervical laminoplasty may be incorrect positioning of neck during surgery, spinal cord edema, reclosure of the laminae, and reperfusion of the decompressed spinal cord based on reports in the literature.[1,3] Though the incidence of acute neurologic deterioration after spine surgery is very low, this complication is considered as the most serious complication for poor prognosis in perioperative period.[4]

In the present case, the patient suffered obvious reactive swelling of the spinal cord after the first surgery, resulting in an acute neurologic deterioration. We hypothesize that the overactive autoimmune reaction induced by AS and limited indirect decompression together contribute to the adverse outcome. AS is a rheumatoid disease of the axial skeletal system which involves extensive soft tissue inflammatory reaction.[8] Though HLA-B27 was negative, the patient was still dignosed with AS for the histroy of lower back pain and typical radiographic evidence of sacroiliitis and bamboo spine changes. These extreme autoimmune inflammatory responses are the basic pathologic changes of AS.[9] The spine deformity and related neurologic dysfunction in AS may necessitate surgical treatment.[10] However, there are no studies reporting the acute neurologic deterioration correlated with AS after laminoplasty.

We speculate an excessive inflammatory reaction and ischemia reperfusion after the surgery may have contributed to the reported complication. It is hard to verify the exact role of AS in this complication. Several signs still suggested that AS appears to be a significant mediating factor. Previous studies have confirmed that AS has the potential to result more serious systematic and local inflammatory reaction after surgery. Reikeras et al[11,12] reported that in patients with AS, the surgery may add a

multifaceted interplay of inflammatory reactions which induce a prolonged and painful postoperative period, likely related to the inflammatory nature of AS. They noticed that the musculoskeletal surgery may induce significantly increase of systemic inflammatory factors in patients with AS, especially in local site where the surgery was performed. In our case, though the ESR and CRP were normal before the surgery, AS may still have the potential to active again under trauma stress. Meanwhile, the spinal cord decompression was associated with reperfusion and edema, particularly at C2/C3 level, and progressed to fill the spinal canal space expanded by the opened laminae, resulting in secondary spinal cord compression and elevated spinal cord tension. The surgical finding and emergency CT supported our views. The dura mater of the cervical spine bulged out of the spinal canal once we removed the mini-plates, indicating elevated tension of the spinal cord. The emergency CT scan documented substantial swelling of the spinal cord, and spinal cord compression at the level of C2/C3. They both indicated the serious inflammatory reaction the spinal cord suffered. Surgeons should be fully aware of the influence of autoimmune disease on surgery result and be prepared for patients with autoimmune disease with regards to the increased potential for overactive inflammatory responses.

Limited decompression in the first surgery may be another factor in the severe complications. Cervical laminoplasty is an indirect decompression method via lifting the lamina to enlarge the spinal canal space which may allow the spinal cord to shift away from the anterior compression. It has advantages including preserving the posterior structures of the cervical spine, decreased postoperative spinal deformities, such as kyphosis or cervical spinal instability, preserving cervical movement, and avoids complications associated with multilevel corpectomy and multilevel laminectomy like nonfusion, spinal deformity.[13,14] It is a well popular posterior cervical surgery to treat multilevel cervical spinal canal stenosis for different causes. This patient suffered cervical spinal canal stenosis from C3 to C7, and we firstly chose cervical laminoplasty for this patient to gain a multilevel decompression. Considering the compression at the C2/C3 level, C2 semilaminectomy was also planned. However, the result suggested that the first treatment plan may not have been enough for this patient considering the large anterior compression mass at the C2/C3 level and the potential severe decompression reaction due to AS. The Pavlov value was 0.19 and occupy ratio was 74% at C3 level. Cervical laminoplasty is an indirect decompression method and is limited for large anterior compression masses. An anterior-posterior surgery or a longer decompression segment involved C2 may be advisable for the initial surgery for this type of patient. Surgeons should be careful to choose cervical laminoplsty facing large anterior compression masses.

It is critically important to respond quickly to acute neurologic deterioration or spinal cord injury.[4] Ahn and Fehlings[1] concluded that the timely recognition was the key to minimizing neurologic dysfunction. They suggest that multimodal neuromonitoring during surgery and regular spinal cord neurologic testing after surgery may help to rapidly identify severe complications. In the present case, several methods were used to quickly diagnose and determine the potential etiology, including spinal cord evoked potential monitoring, regular neurologic inquiry, postsurgical examination, and an emergency CT scan.

Once diagnosed, the causes of acute neurologic deterioration should be addressed as early as possible. Seichi et al[3] reported that 3 patients who had neurologic deterioration after laminoplasty due to lamina reclosure gained slight improvement of symptoms by immediate additional laminectomy. In the present case, the second emergency surgery was arranged immediately. Considering the long compression segments and the need for timely decompression, a posterior surgery along the original approach, but not an anterior approach, was performed. The laminae from C2 to C5 were removed to ensure adequate decompression. The MRIs after the second surgery showed a thorough decompression from C2 to C7, and methylprednisolone was used to decrease postsurgical spinal edema. Methylprednisolone was recommended due to evidence of spinal cord injury and/or a moderate-to-high risk of edema in the cord, as it has a neuroprotective effect by minimizing secondary axonal injury.[1,10] Fortunately, muscle strength recovered to nearly preoperative state 4 days later, and neurologic function was improved at 6-month follow-up.

In conclusion, we reported a rare, acute neurologic deterioration which have been mediated by a pathologic AS reaction following cervical laminoplasty. The potential excessive postsurgical autoimmune response in the patients with rheumatoid diseases should be carefully considered in spinal surgery. Sufficient decompression should always be verified to ensure optimal recovery of neurologic function. Early diagnosis and management is critical in addressing acute neurologic deterioration.

## Author contributions

**Conceptualization:** Hao Liu.
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**Investigation:** Hua Chen, Yuxiao Deng.
**Supervision:** Hao Liu.
**Validation:** Hao Liu.
**Writing – original draft:** Hua Chen.
**Writing – review & editing:** Hao Liu.

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