



Case Report

Multiple Steroid-Induced Vertebral Fracture With Paraparesis Associated With Wegener's Granulomatosis Treated With Posterior Spinal Instrumentation

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Abstract

We report a rare case of multiple steroid-induced vertebral fractures associated with Wegener's granulomatosis. The patient had been administered prednisolone (20 mg/day) for 7 years. He visited us because of severe back pain after a slight fall. Motor weakness was detected below the L1 level. Radiographs and MRI of the spine revealed burst fractures of T5-6, and compression fractures of T9-10, T12, and L3-4 with steroid-induced osteoporosis. At the level of T5-6, the spinal cord was compressed. Conservative therapy had no effect. Posterior spinal fusion using a Hartshill Rectangular Rod and sublaminar wire without decompression was performed. Postoperatively, the back pain disappeared and his neurological deficit and activity of daily living ameliorated.

Keywords: Steroid therapy, vertebral fracture, surgical treatment, Wegener's granulomatosis, thoracic spine, osteoporosis, kyphosis

Posterior Spinal Enstrüman ile Tedavi Edilen Wegener Granülomatozisin Eşlik Ettiği Steroid Tedavisine Bağlı Çoğul Vertebra Fraktürü ve Paraparezi

Özet

Wegener granülomatozu olan ve steroide-bağlı vertebral kırıklı ender bir olgu sunulmaktadır. Hasta 7 yıldan beri prednizolon (20mg/gün) almaktadır. Basit bir düşme sonucu gelişen şiddetli bel ağrısı nedeniyle olgu tarafımızdan görüldü. L1 seviyesi altında motor kuvvetsizlik tesbit edildi. Omurganın radyolojik ve MRG tetkiklerinde T5-6 patlama kırığı yanı sıra T9-10, T12 ve L3-4 seviyelerinde steroide bağlı osteoporotik çökme kırıkları saptandı. T5-6 seviyesinde spinal kord bası altında idi. Konservatif tedavinin bir faydası olmadı. Hartshill Rectangular Rod ve sublaminar tel kullanılarak ve dekompresyon yapılmadan posterior füzyon uygulandı. Ameliyat sonrası hastanın sırt ağrısı geçti, nörolojik bulgularında ve günlük aktivitesinde belirgin düzelme gözlemlendi.

Anahtar Kelimeler: Steroid tedavi, vertebral kırık, cerrahi tedavi, Wegener granülomatozu, dorsal omurga, osteoporoz, kifoz

INTRODUCTION

Wegener's granulomatosis (WG) is a necrotizing, granulomatous vasculitis that has a clinical tendency of involvement of the upper airways, lungs, and kidneys⁽⁶⁾. The prevalence of WG in the United States

has been estimated as 3 per 100,000 with a male-to-female ratio of 1:1⁽¹⁴⁾. Until the introduction of therapeutic regimens with cyclophosphamide (CYC) and glucocorticoids, WG was uniformly fatal⁽⁶⁾. At present, both morbidity and

mortality of patients with WG are influenced by their response to treatment and the degree of therapeutic toxicity⁽⁶⁾.

Among the several side effects of CYC and glucocorticoids in patients with WG⁽³⁾, clinicians have focused on serious infectious diseases due to their close relation to patient mortality^(3,5,7). However, less attention has been paid to secondary osteoporosis in these patients⁽³⁾. Here, we describe a case of WG associated with multiple vertebral fracture and steroid-induced severe osteoporosis, which was resolved successfully by surgery.

CASE PRESENTATION

A 41-year-old man was referred to our clinic because of severe back pain. His clinical history began in 1995 when he consulted an otolaryngologist because of gradual onset of deafness. He was diagnosed as having Wegener's granulomatosis. Since then, he has received oral steroid treatment (prednisolone, 20 mg/day). In 1998, the patient suffered avascular necrosis of the left femoral head induced by steroid treatment, and he underwent femoral head prosthetic replacement. In 2003, the patient developed severe back pain and motor weakness in the lower extremities after a slight fall. At that time, a minor compression fracture of T9 was noted. Conservative treatment for 2 months reduced the symptoms. In this year, the patient underwent right femoral head prosthetic replacement for right avascular necrosis of the femoral head. He then underwent femoral head prosthetic replacement. The back pain became gradually worse, eventually resulting in an inability to walk. In addition, motor weakness below the level of L1 (level 3 to

4 as determined by manual muscle testing), sensory disturbance in the associated area, and paresthesia appeared in both lower extremities. Deep tendon reflexes in the lower extremities were exaggerated. Plain radiographs of the thoracic and lumbar spine revealed burst fractures of T5 and 6 and compression fractures of T9, T10, T12, L3, L4, and L5 in the osteoporotic spine in a kyphotic shape. (Figure 1A) Magnetic resonance imaging (MRI, T2-weighted) showed collapse of T5 and T6 with moderate spinal cord compression. (Figure 1B) An area of high signal intensity in the spinal cord at T 6/7 was noted. Conservative treatments were not effective, and the patient could not leave his bed. Therefore, he underwent posterior spinal fusion with a Hartshill rectangular rod® (HRR) (Surgicraft, Redditch, United Kingdom)⁽¹⁾, and sublaminar wires (SLW) without decompression between T2 and T10. During the operation, his thoracic spine was controlled to decrease its kyphosis by the postural reduction.

Postoperative plain X-ray examination indicated a decrease in the Cobb angle between T2 and T10 from 17° to 14°, and the patient's back pain disappeared. With recovery of motor function below the level of L1 (level 4 as determined by manual muscle testing), he regained ambulatory ability in a wheelchair. However, the paresthesia of both lower extremities did not show complete amelioration. After a 2-year follow-up, the patient is free from pain and able to walk using a walker. Plain X-ray examination of his spine indicated no subsequent compression fracture or loss of sagittal alignment in the thoracic spine (Figure 2). The patient is satisfied with the surgical outcome.



Figure 1A: Preoperative lateral radiograph showing burst fractures of T5 and T6, and a compression fracture of T9, T10, T12, and L3, L4, and 5. Total alignment was pronounced kyphosis.



Figure 1B: Magnetic resonance images (T2-weighted) showing collapse of T5 and T6 with moderate spinal cord compression. Note the high signal intensity area in the spinal cord at T6/7.

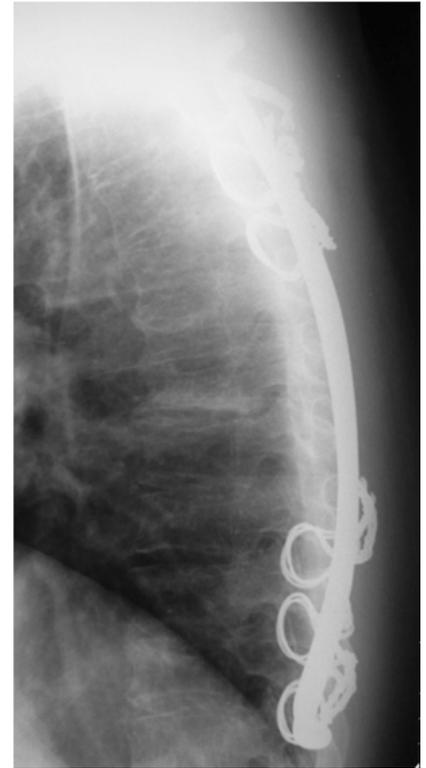


Figure 2: Lateral radiographs taken 2 years after the operation. No compression fracture was detected.

DISCUSSION

The development of therapeutic regimens, including CYC, glucocorticoids,⁽¹⁾ and methotrexate⁽²⁾, reduced the high mortality rates of patients with WG⁽¹⁴⁾. However, it is still necessary to determine how best to deal with the side effects of these treatments. In the field of orthopedics, corticosteroid therapy has been reported to be associated with a dose-related increase in risk of bone mineral density loss, making patients susceptible to fractures⁽¹³⁾. To our knowledge, there have been no

previous reports of multiple vertebral fractures due to steroid-induced osteoporosis in patients with Wegener's granulomatosis, as in the present case.

In the present case, osteoporosis was so severe that the patient's severe pain was resistant to conventional conservative therapies. The patient also showed neurological deficits. However, it was necessary to continue steroid treatment to manage WG. Furthermore, long-term bed rest appeared to exacerbate his general

condition. Therefore, surgical treatment was indicated for this patient.

The indication of surgical treatment in patients with multiple vertebral fracture due to osteoporosis is still controversial⁽¹¹⁾ due to poor bone quality for spinal instrumentation⁽¹²⁾, difficulty in selecting the fusion area, poor general condition in the majority of cases, and possible peri- and postoperative complications⁽⁴⁾. First, we felt it necessary to reinforce the thoracic spine, otherwise successive collapse of the vertebral bodies would have compressed the spinal cord. Assuming that pedicle screws would not have sufficient fixation due to osteoporosis⁽¹²⁾, segmental spinal fixation⁽⁴⁾ using HRR+SLW⁽¹⁾ was indicated for this patient. Taking his general condition into account, anterior decompression was not performed. As the compression to the spinal cord was from the front, laminectomy was not performed. Consequently, the operation was effective both in reducing the pain and in ameliorating the neurological deficit. We speculated that the amelioration of the neurological deficit in this case was due to two factors. First, simply reinforcing the spine with the rods reduced the dynamic factor of spinal cord compression⁽¹⁰⁾. Second, even though the change in Cobb angle was slight, the decrease in kyphotic angle which was obtained by postural reduction in a prone position and was maintained by the posterior instrumentation relieved anterior compression of the spinal cord by the bony fragment⁽⁹⁾.

When other surgical method for reinforcing the thoracic spine of the present patient is reconsidered, vertebroplasty could have been an additional or alternative choice. Because there were severe osteoporosis and multiple levels compromise, the vertebroplasty might have been better performed with an augmentation using pedicle screw fixation⁽⁸⁾ or sublaminar wire fixation.

Anterior decompression⁽⁴⁾ for spinal cord compression can also be performed as an additional choice with regard to the postoperative clinical course. As the clinical outcome of the patient has been uneventful and treatment immediately relieved the pain and later improved the patient's motor weakness, no further surgery has been performed. However, as the patient must continue to take steroids for the control of WG, further periodical observation is necessary.

CONCLUSION

A case of multilevel thoracic vertebral fracture due to steroid therapy for Wegener's granulomatosis was reported. Posterior fusion using a Hartshill Rectangular Rod and sublaminar wire without decompression provided relief of severe pain and partial recovery in activity of daily living.

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Received by: 11 July 2007

Revised by: 12 December 2007

Accepted : 28 December 2007

The Online Journal of Neurological Sciences (Turkish) 1984-2008

This e-journal is run by Ege University Faculty of Medicine,

Dept. of Neurological Surgery, Bornova, Izmir-35100TR

as part of the Ege Neurological Surgery World Wide Web service.

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Journal of Neurological Sciences (Turkish)

Abbr: J. Neurol. Sci.[Turk]

ISSNe 1302-1664

REFERENCES

1. Dove J. Internal fixation of the lumbar spine. The Hartshill rectangle. *Clin Orthop*. 1986; 135-40.
2. Hoffman GS, Leavitt RY, Kerr GS, et al. Treatment of glucocorticoid-resistant or relapsing Takayasu arteritis with methotrexate. *Arthritis Rheum*. 1994; 37:578-82.
3. Hoffman GS, Kerr GS, Leavitt RY, et al. Wegener granulomatosis: an analysis of 158 patients. *Ann Intern Med*. 1992; 116:488-98.
4. Hu SS. Internal fixation in the osteoporotic spine. *Spine*. 1997; 22:43S-8S.
5. Krafcik SS, Covin RB, Lynch JP, et al. Wegener's granulomatosis in the elderly. *Chest*. 1996; 109:430-7.
6. Langford CA. Wegener granulomatosis. *Am J Med Sci*. 2001; 321:76-82.
7. Mahr A, Girard T, Agher R, et al. Analysis of factors predictive of survival based on 49 patients with systemic Wegener's granulomatosis and prospective follow-up. *Rheumatology (Oxford)*. 2001; 40:492-8.
8. Matsuyama Y, Yoshihara H, Tsuji T, et al. Vertebral reconstruction with biodegradable calcium phosphate cement in the treatment of osteoporotic vertebral compression fracture using instrumentation. *J Spinal Disord Tech*. 2004; 17:291-6.
9. Matsuyama Y, Yoshihara H, Tsuji T, et al. Surgical outcome of ossification of the posterior longitudinal ligament (OPLL) of the thoracic spine: implication of the type of ossification and surgical options. *J Spinal Disord Tech*. 2005; 18:492-7.
10. Olsson SE. The dynamic factor in spinal cord compression; a study on dogs with special reference to cervical disc protrusions. *J Neurosurg*. 1958; 15:308-21.
11. Phillips FM. Minimally invasive treatments of osteoporotic vertebral compression fractures. *Spine*. 2003; 28:S45-53.
12. Soshi S, Shiba R, Kondo H, et al. An experimental study on transpedicular screw fixation in relation to osteoporosis of the lumbar spine. *Spine*. 1991; 16:1335-41.
13. van Staa TP, Leufkens HG, Cooper C. The epidemiology of corticosteroid-induced osteoporosis: a meta-analysis. *Osteoporos Int*. 2002; 13:777-87.
14. Yi ES, Colby TV. Wegener's granulomatosis. *Semin Diagn Pathol*. 2001; 18:34-46.