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Case Report

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Cervical Spinal Fracture Caused by Untreated Tourette Syndrome: A Case Report

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Conflict of Interest
The authors have no financial conflicts of interest.

ABSTRACT

Cervical myelopathy can occur in Tourette syndrome patients with severe motor tics showing repetitive and violent neck movements. However, motor tics causing spinal fractures have been rarely reported. A 15-year-old girl presented at our clinic, complaining of recent development of motor weakness of all 4 extremities. She had untreated motor tics involving the neck. Computed tomography and magnetic resonance imaging findings suggested cervical spinal fractures and myelopathy. After diagnosing of Tourette syndrome, medical and psychologic therapies were started. Her motor tics were well controlled, and no complications in the patient's daily life were observed later. Cervical radiography taken at a 9-month follow-up showed bony healings of the fractured cervical spines. Uncontrolled severe motor tics may cause spinal fractures. Conservative treatments would suffice for proper control of these tics and stabilize the spine, and considered as initial treatment in patients with Tourette syndrome.

Keywords: Tic disorders; Tourette syndrome; Cervical vertebrae; Myelopathy; Spinal fractures

INTRODUCTION

Tourette syndrome is a relatively common neuropsychiatric disease in young age, and manifests many types of motor and vocal tics with variable severity.[2] Although rare, severe motor tics with repetitive and violent neck movements can cause various degrees of cervical myelopathy or radicular symptoms.[3,6,8,9,11,13] Several studies had suggested that spondylotic changes and dynamic instability of the cervical spine caused and exacerbated by excessive neck movements is the myelopathic mechanism.[6,8,11] However, in contrast to standard cervical spondylotic myelopathy, the spinal cord damage is frequently seen on magnetic resonance imaging (MRI) in the upper cervical spine in myelopathies related to movement disorders.[12] In addition, intensity and/or extent of the cord signal change is often severe,[1,5,12] and sometimes it shows in various and unusual locations of the spinal cord.[6] These various findings cannot be explained in a single, simple explanation, and it would be reasonable to think that motion related complex pathophysiologic mechanisms underlie in myelopathies related movement disorders.

We encountered a patient with untreated Tourette syndrome and recently developed weakness of 4 extremities. The patient had multiple cervical vertebral fractures without any noticeable trauma events. To the best of our knowledge, this is the first report describing severe motor tics involving the neck causing vertebral fractures.

CASE REPORT

A 15-year-old girl presented at our clinic, complaining of recent development of motor weakness of all 4 extremities that started approximately 3 weeks prior. The patient did not complain neck pain, and there was no history of trauma. Neurologic examination revealed a good grade of muscle power of all of 4 extremities, but the patient complained subjective weakness, more on right arm and leg. But left hemibody sensory changes, hyperreflexic deep tendon reflexes, positive Babinski reflexes and ankle clonuses indicated an upper motor neuron lesion.

During her hospital stay for the diagnostic work-up, we learned a new fact that she had been suffering a tic disorder that began when she was approximately 5–6 years old. Vocal tic was first, and approximately 2 years before the admission, a motor tic emerged. She repeatedly hyper-flexed and extended the neck many times during a day until a cracking sound emerged from her neck. Her mother said that this tic movement repeated at least 20–30 times an hour, especially when she was watching television at home. She had consulted doctors for the treatment. However, she soon quit treatment because the medication made her sleepy and she had trouble concentrating. The motor tic in her neck had been untreated for 2 years.

Cervical spine radiography and computed tomography showed several anterior-wedge deformed subaxial vertebral bodies with endplate irregularities, and small bony fragments detached from anterior vertebral bodies were observed (**FIGURES 1** & **2**). No mechanical instability was observed (**FIGURE 1B & C**). MRI showed bone marrow edema from C4 to C7 vertebral bodies (**FIGURE 3**). These findings suggested acute to subacute stage vertebral body fractures. T2-weighted sagittal MRI suggested the presence of a high cord signal change at the

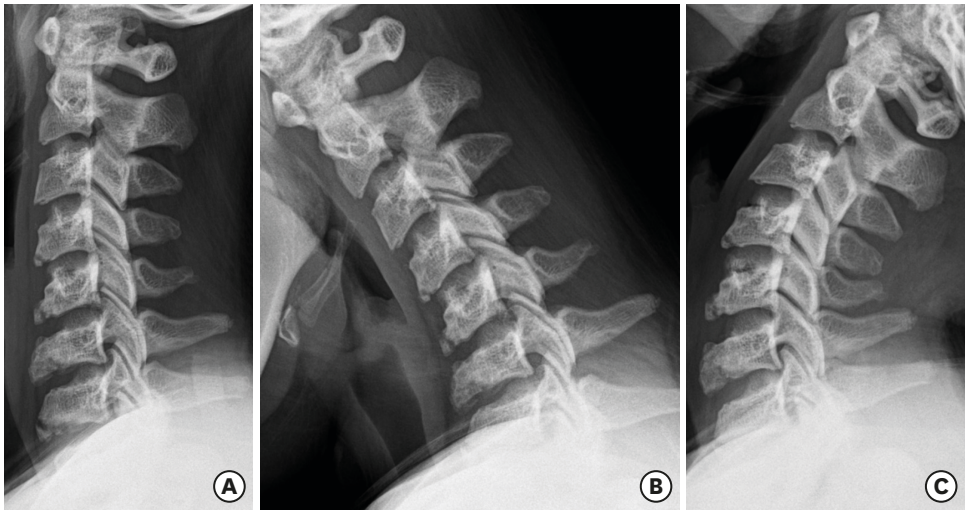

FIGURE 1. Initial C-spine (A) neutral, (B) flexion and (C) extension lateral views. Anterior-wedge deformed vertebral bodies with endplate irregularities and osteophytes at C4 to C7 are shown. No instability of C-spine is observed.

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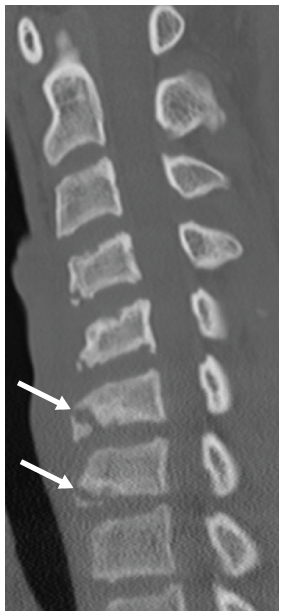

FIGURE 2. On computed tomography, small bony fragments (white arrows) are detached from lower endplate of the anterior vertebral bodies in C4 to C7.

C6 level, but this finding could not be confirmed by axial scan of that level (**FIGURE 3B & D**). T1 enhance MRI was taken to confirm any presenting cord pathology, no enhancement was observed on the spinal cord. Since non-traumatic vertebral fractures in a young patient are hard to expect, we conducted more tests to ascertain signs of systemic metabolic disorders. All laboratory test results were within the normal limits. Her bone mineral density was normal. On the bone scan, only the low cervical spines showed increased uptakes which suggested recent fractures (**FIGURE 4**) By summarizing the results, we came to the conclusion that the motor tic involved the neck had caused the cervical myelopathy and fractures.

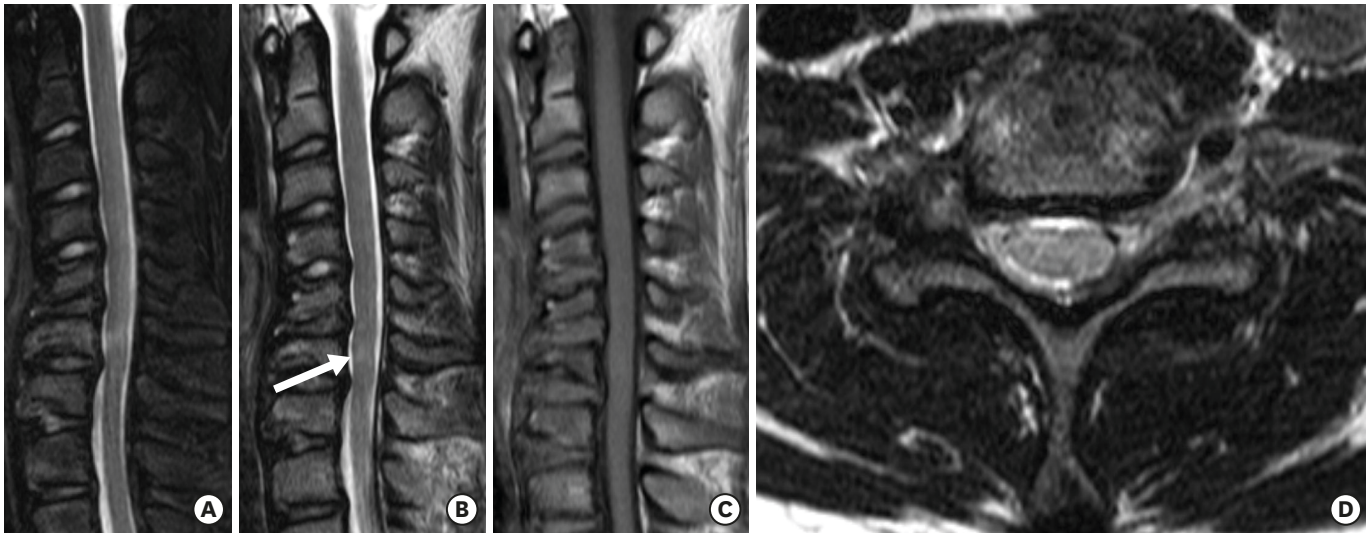

FIGURE 3. (A) Sagittal T2-WI with fat saturation, (B) T2-WI, (C) T1-WI, and (D) axial T2-WI of magnetic resonance imaging. (A-C) Small bone fragments with T2 iso- to high signal, and T1 low signal change are shown in C4 to C7 vertebral bodies, suggesting acute to subacute stage of fractures. (B, D) Subtle high signal change (white arrow) in the spinal cord of C6 body level, possibly early stage of myelopathy. However, this finding is not definite on axial image on that level. WI: weighted image.

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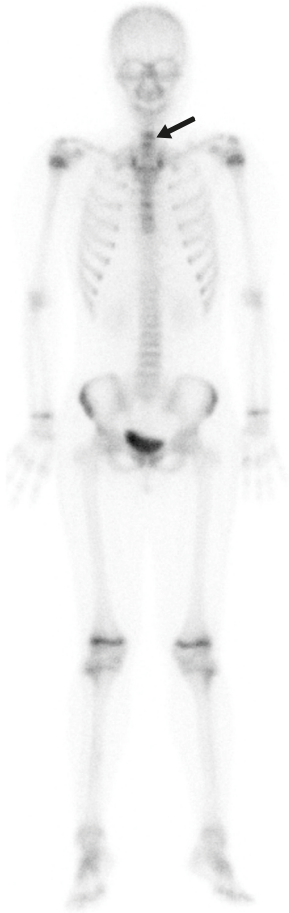

FIGURE 4. Whole body bone scan showed increased uptakes at the lower cervical spine (black arrow), suggesting recent fractures. No other abnormal uptakes were observed.

We thought that the cervical spine would be stable with proper control of the motor tic. A Philadelphia neck brace was applied, and we consulted the psychiatric department for proper management of the tic disorder. Tourette syndrome was diagnosed. With patient education, supportive psychotherapy and medication, the motor tic was well controlled. No other medication related to the myelopathy such as steroids was used. For 9 months of follow-up, she only complained of a subjective decrease in her right leg motor power during strenuous exercises. No complications in her daily life were observed. Follow-up cervical spine radiography confirmed bone healing of the lower cervical spines (**FIGURE 5**).

DISCUSSION

Through reports and experiences with athetoid-type cerebral palsy,[7] it is well known that movement disorders exhibiting violent repetitive movements involving the head and neck may cause cervical myelopathy. Similar reports regarding Tourette syndrome also present in the literature.[3,6,8,9,11,13] The pathophysiology of cervical myelopathy caused by motor tics of the neck is not clearly elucidated. Compression against the spinal cord by canal stenosis from excessive spondylotic changes combined with severe dynamic instability of the spine caused by constant tic movements is an acceptable explanation in adult patients showing early

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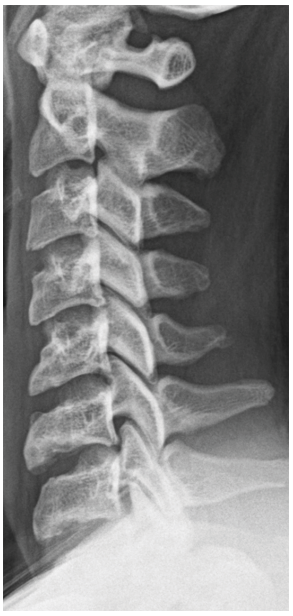

FIGURE 5. The 9-month follow-up C-spine lateral image shows no fracture line and completely union of previous fracture fragments in C4 to C7 vertebral bodies.

degenerative changes.[8] Some authors had suggested that direct compression or excessive stretching of the spinal cord by repetitive extreme posture of the neck may damage the cord.[6,8] Acute myelopathy caused by jeopardizing spinal arteries had also been suggested in a few cases.[8,13]

On the other hand, evidence of motor tics causing spine fractures were scarce. In our best knowledge, there was only one case report of stress fractures of both peroneal bone caused by complex motor tics.[4] Lu et al.[10] reported a positive correlation between Tourette syndrome and risk of bone fractures, but this correlation was associated with self-injuries behavior, not with repetitive movements of motor tics. Combined with the fact that there was no trauma history and the patient did not complain of any pain in the neck, presence of unusual systemic metabolic disorders which might make the cervical vertebrae weak was suspected initially.

In reviewing the present case, the diagnosis of cervical myelopathy was not confirmatory. Radiologic findings did not show any compressive lesion against neural structures or dynamic instability. Subtle cord signal change at C6 level was suspected on MRI. However, this finding was only confined to one imaging sequence. Despite these uncertainties, we think it is possible that the patient had myelopathy at C6 level. As mentioned previously, the cord signal change on MRI in patients with movement disorders is quiet different from that of standard cervical spondylotic myelopathy. Most important factor for such difference seems to be the motion of involved spine. For example, a propensity of upper cervical cord damage owes to rotating movements caused by movement disorders.[12] Where severe, excessive motions damage the spinal stability, myelopathy ensue. In this case, the maximal motion stress induced by repetitive flexion and extension tic movements seem to present at C6 level. C5–6 and C6–7 motion segments are where most active flexion and extension movements of cervical spine occur. In addition, 2 radiologic findings in this patient support this assumption. First, the C6 vertebral body had the largest anterior fractured bony fragment (**FIGURE 2**). Second, most anterior wedge deformed vertebrae were C5 to C7, and C6 is

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located at the center of them. Although any dynamic instability was not seen, fractured spines might have gone over the limits of physiologic range of motion, resulting in myelopathy. This hypothesis provides a plausible explanation what had happened. However, we should be very careful in accepting this since the evidences are very weak.

Fortunately, the patient's motor tics were well controlled after starting medical and supportive psychiatric treatments for Tourette syndrome, and the fractured cervical vertebrae completely healed approximately after 9 months later. We reconfirm that the fractures were truly caused by the tics.

CONCLUSION

Patients with a severe motor tic may cause cervical fractures with or without myelopathy. In cases of Tourette syndrome lacking spinal instability or severe neurologic deficits, conservative treatments with a proper motor tic control may suffice.

REFERENCES

1. Aguirregomozcorta M, Pagonabarraga J, Diaz-Manera J, Pascual-Sedano B, Gironell A, Kulisevsky J. Efficacy of botulinum toxin in severe Tourette syndrome with dystonic tics involving the neck. **Parkinsonism Relat Disord 14**:443-445, 2008
PUBMED | CROSSREF
2. Bloch MH, Leckman JF. Clinical course of Tourette syndrome. **J Psychosom Res 67**:497-501, 2009
PUBMED | CROSSREF
3. Dobbs M, Berger JR. Cervical myelopathy secondary to violent tics of Tourette's syndrome. **Neurology 60**:1862-1863, 2003
PUBMED | CROSSREF
4. Fusco C, Bertani G, Caricati G, Della Giustina E. Stress fracture of the peroneal bone secondary to a complex tic. **Brain Dev 28**:52-54, 2006
PUBMED | CROSSREF
5. Huasen B, McCreary R, Evans J, Potter G, Silverdale M. Cervical myelopathy secondary to Tourette's syndrome managed by urgent deep brain stimulation. **Mov Disord 29**:452-453, 2014
PUBMED | CROSSREF
6. Isaacs JD, Adams M, Lees AJ. Noncompressive myelopathy associated with violent axial tics of Tourette syndrome. **Neurology 74**:697-698, 2010
PUBMED | CROSSREF
7. Kim KN, Ahn PG, Ryu MJ, Shin DA, Yi S, Yoon DH, et al. Long-term surgical outcomes of cervical myelopathy with athetoid cerebral palsy. **Eur Spine J 23**:1464-1471, 2014
PUBMED | CROSSREF
8. Ko DY, Kim SK, Chae JH, Wang KC, Phi JH. Cervical spondylotic myelopathy caused by violent motor tics in a child with Tourette syndrome. **Childs Nerv Syst 29**:317-321, 2013
PUBMED | CROSSREF
9. Krauss JK, Jankovic J. Severe motor tics causing cervical myelopathy in Tourette's syndrome. **Mov Disord 11**:563-566, 1996
PUBMED | CROSSREF
10. Lu YY, Wang MY, Wei IH, Lin CC, Huang CC. Tourette syndrome increases risk of bone fractures: a population-based cohort study. **Eur Child Adolesc Psychiatry 26**:531-539, 2017
PUBMED | CROSSREF
11. Miyashita T, Yamazaki M, Okawa A, Yoneda M, Aiba A, Koda M, et al. Multiple neck operations in a patient with severe motor tics because of Tourette's syndrome: a case report. **J Med Case Rep 6**:223, 2012
PUBMED | CROSSREF

https://doi.org/10.13004/kjnt.2020.16.e23

12. Muroi A, Matsumura A, Asakawa H, Enomoto T, Iwasaki N, Nose T. Myelopathy caused by tics in an adolescent, associated with T2 signal intensity changes of the spinal cord. **Childs Nerv Syst 18**:191-194, 2002
PUBMED | CROSSREF

13. Patterson AL, Choudhri AF, Igarashi M, McVicar K, Shah N, Morgan R. Severe neurological complications associated with Tourette syndrome. **Pediatr Neurol 61**:99-106, 2016
PUBMED | CROSSREF

https://doi.org/10.13004/kjnt.2020.16.e23