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A New Concept for Making Decisions Regarding the Surgical Approach for Cervical Ossification of the Posterior Longitudinal Ligament

The K-Line

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Study Design. To report a new index, the K-line, for deciding the surgical approach for cervical ossification of the posterior longitudinal ligament (OPLL).

Objective. To analyze the correlation between the K-line-based classification of cervical OPLL patients and their surgical outcome.

Summary of Background Data. Previous studies showed that kyphotic alignment of the cervical spine and a large OPLL are major factors causing poor surgical outcome after laminoplasty for cervical OPLL patients. However, no report has evaluated these 2 factors in 1 parameter.

Methods. The K-line was defined as a line that connects the midpoints of the spinal canal at C2 and C7. Twenty-seven patients who had cervical OPLL and underwent posterior decompression surgery were classified into 2 groups according to their K-line classification. OPLL did not exceed the K-line in the K-line (+) group and did exceed it in the K-line (−) group. By intraoperative ultrasonography, we evaluated the posterior shift of the spinal cord after the posterior decompression procedure. The Japanese Orthopedic Association scores before surgery and 1 year after surgery were evaluated, and the recovery rate was calculated.

Results. Eight patients were classified as K-line (−), and 19 patients were classified as K-line (+). The mean recovery rate was 13.9% in the K-line (−) group and 66.0% in the K-line (+) group ($P < 0.01$). Ultrasonography showed that the posterior shift of the spinal cord was insufficient in the K-line (−) group.

Conclusion. The present results demonstrate that a sufficient posterior shift of the spinal cord and neurologic improvement will not be obtained after posterior decompression surgery in the K-line (−) group. Our new index, the K-line, is a simple and practical tool for making decisions regarding the surgical approach for cervical OPLL patients.

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Key words: K-line, surgical approach, ossification of posterior longitudinal ligament, cervical myelopathy, laminoplasty. **Spine 2008;33:E990–E993**

Regarding the factors causing poor surgical outcomes after laminoplasty for cervical ossification of the posterior longitudinal ligament (OPLL), previous reports have described 2: (1) the alignment of the cervical spine is kyphotic,[1] and (2) the size of the OPLL is large.[2,3] Previous studies evaluated these 2 factors independently, and, to the best of our knowledge, no report has analyzed them in 1 parameter. In the present study, we proposed a new index that can evaluate the cervical alignment and the OPLL size in 1 parameter. We named this index the K-line. The "K" stands for "kyphosis." According to the K-line, we classified cervical OPLL patients who underwent posterior decompression surgery, and evaluated the relationship between their surgical outcome and the K-line-based classification. In addition, we used intraoperative ultrasonography (US) to evaluate the posterior shift of the spinal cord from the OPLL and analyzed the relationship between the decompression status of the spinal cord and the K-line classification.

Materials and Methods

Patients

From May 1990 through December 2005, 27 patients with cervical myelopathy due to OPLL underwent posterior decompression surgery. The patients included 23 males and 4 females. Their mean age at surgery was 63.3 years (range, 46–81 years). The surgical method included cervical enlargement laminoplasty in 19 patients and posterior decompression with instrumented fusion in 8 patients. Our cervical enlargement laminoplasty consisted of a C3 to C7 *en bloc* laminoplasty (Itoh's method).[4,5] From April 2003, when cervical OPLL patients have massive OPLL and evident intersegmental mobility at the cord compression level, we performed posterior instrumented fusion of the cervical spine using a lateral mass and rod system associated with laminectomy or laminoplasty as described previously.[5] All patients had undergone follow-up evaluation for a period of 1 year or longer after surgery.

Clinical and Radiographic Assessments

The Japanese Orthopedic Association scoring system was used to evaluate the severity of cervical myelopathy.[5] The Japanese Orthopedic Association scores before surgery and 1 year after surgery were evaluated, and the recovery rate was calculated.[5]

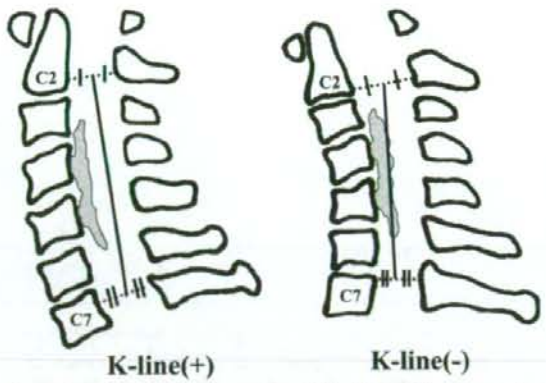

Figure 1. Schematic drawings of the "K-line." The K-line is a straight line that connects the midpoints of the spinal canal at C2 and C7 on the lateral cervical radiographs. Patients with cervical ossification of the posterior longitudinal ligament (OPLL) are divided into 2 groups according to the K-line. OPLL does not exceed the K-line in the K-line (+) group and does exceed it in the K-line (−) group.

Before surgery, the cervical lordotic angle (C2–C7 angle) was measured on lateral radiographs.[5] The occupation ratio of the ossified mass at the most stenotic level of the spinal canal was examined with computed tomography (CT) and defined as follows: OPLL occupation ratio = (thickness of OPLL/anteroposterior diameter of the bony spinal canal) × 100.[5]

Definition of K-Line

To draw the K-line, we used principally the lateral view of the cervical radiograph in the neutral position. In cases whose C7 vertebrae were occluded by the shadow of the patients' shoulders, we evaluated the midsagittal view of the T2-weighted magnetic resonance image. We first decided the midpoints of the spinal canal at C2 and C7 and then connected them (Figure 1). According to the K-line, OPLL cases were divided into 2 groups: a K-line (+) group and a K-line (−) group (Figure 1). In the K-line (+) group, the OPLL did not exceed the K-line and stayed within the ventral area of the K-line. Because there is a space between the K-line and OPLL, we named this group "plus" (Figure 2A). In the K-line (−) group, OPLL exceeded the K-line and had grown beyond the K-line (Figures 2B, C). When the size of OPLL was large, many cases were classified as K-line (−) (Figure 2B). Even though the OPLL size was not large, some OPLL cases were classified as K-line (−) when the alignment of the cervical spine was kyphotic (Figure 2C).

Intraoperative Spinal US

By means of intraoperative US, we recorded the movement of spinal cord by an animation mode and assessed the posterior shift of the spinal cord from the anterior ossified mass. Immediately after the posterior decompression procedure, we evaluated the presence of the subarachnoid space at the ventral side of the spinal cord. We then classified the decompression status into 3 types: a noncontact type, a contact and apart type, and a contact type.[6] In the noncontact type, the spinal cord does not touch the OPLL, and the subarachnoid space can always be seen between the OPLL and the spinal cord. In the contact and apart type, the spinal cord touches and is apart from the OPLL, depending on the pulsation. In the contact type, the spinal cord always touches the OPLL, and no subarachnoid space appears between the OPLL and the spinal cord.

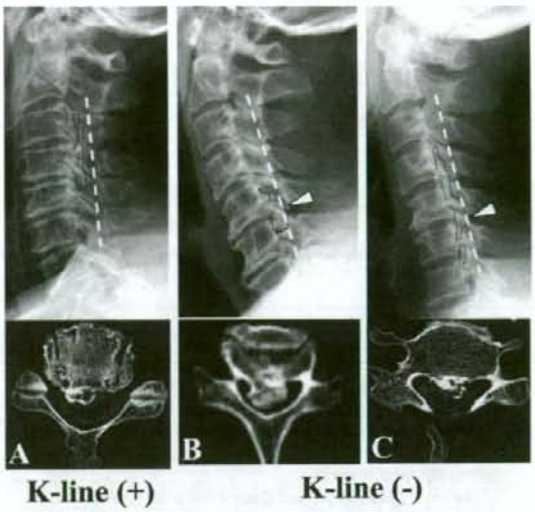

Figure 2. Representative cervical radiographs and computed tomography (CT) images of the K-line (+) (**A**) and the K-line (−) (**B, C**). **A,** The size of OPLL is not large (the occupation ratio of the spinal canal by OPLL is 36.3%, and the OPLL thickness is 4 mm at C4/5), and the OPLL stays at the ventral area of the K-line. **B,** The OPLL size is large (the occupation ratio is 85%, and the thickness is 11 mm at C5/6), and the OPLL extends beyond the K-line. **C,** The OPLL size is intermediate (the occupation ratio is 48.4%, and the thickness is 5 mm at C5/6), but the OPLL exceeds the K-line because the alignment of the cervical spine is kyphotic. Arrowheads in (**B**) and (**C**) indicate the site where the OPLL exceeds the K-line.

Statistical Analysis

Statistical analysis was performed using the Mann-Whitney *U*-test, the Scheffe's F test and Fisher's exact probability test. A $P < 0.05$ was considered statistically significant. Results are presented as the mean ± standard error of the mean.

■ Results

Of the 27 OPLL patients analyzed in the present study, 19 were classified as K-line (+), and 8 were classified as K-line (−). Preoperative and postoperative clinical data are summarized in Table 1. The mean recovery rate was 66.1% in the K-line (+) group and 13.9% in the K-line (−) group. Thus, neurologic improvement after surgery was far better in the K-line (+) group than in the K-line (−) group ($P < 0.01$).

Five patients were classified as the contact type by intraoperative US, 12 patients were classified as the contact and apart type, and 10 patients were classified as the noncontact type. The mean recovery rate was 10.5% in patients of the contact type, 54.2% in patients of the contact and apart type, and 66.5% in patients of the noncontact type. Thus, neurologic improvement in patients of the contact type was inferior to those of the contact and apart type and the noncontact type ($P < 0.05$).

Of the 19 patients in the K-line (+) group, 9 patients were classified as the contact and apart type, and 10 patients were classified as the noncontact type. Of the 8 patients of the K-line (−) group, 5 patients were classi-

Table 1. Clinical Data According to the K-Line Classification

K-Line Group	K-Line (+) (n = 19)	K-Line (−) (n = 8)
JOA score (points)*		
Before surgery	8.9 ± 0.7 (4–14)	7.3 ± 0.7 (4.5–9.5)
After surgery	13.8 ± 0.6 (10–17)	8.9 ± 0.8† (5–12)
Recovery rate (%)*	66.1 ± 5.2 (33.3–100)	13.9 ± 12.2† (−50–52)
Age at surgery (yr)*	60.7 ± 1.9 (46–78)	69.5 ± 3.6 (54–81)
Occupation ratio of OPLL (%)*	52.3 ± 2.9 (28.6–72.7)	63.8 ± 5.0 (45.4–90)
Thickness of OPLL (mm)*	6.7 ± 0.4 (3–9)	8.4 ± 0.6 (6–11)
C2–C7 Lordotic angle (degrees)*	12.4 ± 1.9 (−3–26)	13.7 ± 5.9 (−12–32)
Ultrasonography type (n)		
Contact	0	5
Contact and apart	9	3
Noncontact	10	0

*Values are expressed as mean ± standard error, with the range in parentheses.
†Statistically different from the K-line (+) group ($P < 0.01$).
JOA indicates Japanese Orthopaedic Association.

fied as the contact type and 3 patients were classified as the contact and apart type (Table 1). Thus, the incidence of the contact type was significantly higher in the K-line (−) group ($P < 0.01$). In contrast, the incidence of the noncontact type was significantly higher in the K-line (+) group ($P < 0.05$).

■ Discussion

Batzdorf *et al*[7] analyzed the degree of spinal curvature in patients with cervical spondylotic myelopathy and reported the relation between the degree of curvature and the postoperative clinical outcome after laminectomy. Taking into account their study design, in the present study, we advocated the K-line as an index for deciding the surgical approach for cervical OPLL patients. By using the K-line, we can evaluate the alignment of the cervical spine and the size of OPLL in 1 parameter. In addition, the method of patient classification according to the K-line is simple and practical.

With surgeries from the posterior approach for cervical OPLL patients, we can evaluate the posterior shift of the spinal cord immediately after the posterior decompression procedure by using intraoperative US. Mihara *et al*[8] analyzed the findings of intraoperative spinal US in cervical myelopathy patients who underwent laminoplasty. They reported that better neurologic recovery was obtained in patients in whom the subarachnoid space appeared at the ventral side of the spinal cord. This is consistent with the findings of our present study that, in patients whose intraoperative US findings were the contact type, neurologic improvements after surgery were inferior to those of the contact and apart type and the noncontact type.

When we perform surgeries for cervical OPLL patients, we expect complete decompression of the spinal cord. Thus, the most desirable intraoperative US finding immediately after the posterior decompression procedure is the noncontact type. In the K-line (−) group in the present study, no patient was a noncontact type. This indicates that the posterior shift and the decompression of the spinal cord from the anterior ossified mass will be insufficient in the K-line (−) group. In contrast, in the K-line (+) group, 10 patients were the noncontact type, and no patient was the contact type. This indicates that, in many cases in the K-line (+) group, a sufficient posterior shift decompression of the spinal cord will be expected, even after surgeries from the posterior approach. If we select posterior surgeries for cervical OPLL patients of the K-line (−) group, an adequate posterior shift of the spinal cord will not occur after surgery, and a sufficient neurologic improvement will not be expected. Thus, we suggest that, for patients of the K-line (−) group, selection of surgeries from the posterior approach is not appropriate. For such patients, we recommend anterior decompression surgery as the first choice.

In conclusion, the present results demonstrate that a sufficient posterior shift and decompression of the spinal cord will not be obtained with surgeries from the posterior approach for cervical OPLL patients of the K-line (−) group. Our new index, the K-line, is a simple and practical tool for making decisions regarding the surgical approach for cervical OPLL patients.

■ Key Points

- We proposed a new index, the K-line, which can evaluate cervical alignment and OPLL size in 1 parameter.
- Cervical OPLL patients who underwent posterior decompression surgery were classified into 2 groups: K-line (+) and K-line (−).
- Sufficient posterior shift of the spinal cord and neurologic improvement were not obtained after posterior decompression surgery in the K-line (−) group.
- The K-line is a simple and practical tool for making decisions regarding the surgical approach for cervical OPLL patients.

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Infection-Related Atlantoaxial Subluxation (Grisel Syndrome) in an Adult With Down Syndrome

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Study Design. Case report.

Objective. To report an adult case with Down syndrome, in whom infection-related atlantoaxial subluxation (Grisel syndrome) developed.

Summary of Background Data. Atlantoaxial instability is a common complication in Down syndrome patients; however, there have been limited reports of adult-onset atlantoaxial subluxation with myelopathy. Grisel syndrome has been characterized as a nontraumatic atlantoaxial subluxation associated with pharyngeal infection. It usually affects children, and the subluxation can be successfully reduced by conservative treatments in most cases.

Methods. A 26-year-old man with Down syndrome suffered from retropharyngeal infection, after which his atlantoaxial subluxation was aggravated and myelopathy developed. He was treated with administration of antibiotics and application of a halo-vest.

Results. The conservative treatments failed to reduce the atlantoaxial subluxation. We performed a C1 laminectomy and posterior occipitocervical fusion, which successfully relieved his symptoms.

Conclusion. This experience suggests that when Down syndrome patients have retropharyngeal infection, we should consider the possible aggravation of atlantoaxial instability and development of myelopathy, even if the patient is an adult.

Key words: Down syndrome, atlantoaxial subluxation, infection, Grisel syndrome, adult. **Spine 2008;33: E156–E160**

In patients with Down syndrome, atlantoaxial subluxation (AAS) associated with ligamentous laxity is a major complication with a relatively high incidence, ranging from 12% to 24%.[1–3] Despite such a high incidence of AAS, the development of myelopathy has been reported in only 1% of cases.[4] When Down syndrome patients have congenital skeletal anomalies at the craniovertebral junction (CVJ) such as os odontoideum, the risk of the development of myelopathy increases.[5,6]

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The device(s)/drug(s) that is/are the subject of this manuscript is/are not FDA-approved for this indication and is/are commercially available in the United States.

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Previous reports have shown that the average age of onset of myelopathy due to AAS in Down syndrome patients is 10.5 years.[2] The risk of myelopathy due to AAS is thought to decrease with age. According to Semine *et al*, the laxity of the transverse ligament of the atlas decreases with aging. As a result, atlantoaxial instability often disappears before patients reach adulthood.[1] In actuality, there have been limited reports of adult-onset AAS with myelopathy in Down syndrome patients.[7]

We report an adult case with Down syndrome, in whom infection-related AAS (Grisel syndrome) developed.[8] In the present case, retropharyngeal infection aggravated the AAS, resulting in the development of myelopathy.

Case Reports

Clinical Profile

A 26-year-old man with Down syndrome was admitted to our hospital with complaints of severe neck pain and gait disturbance. Two months before admission, he had felt neck pain without trauma. Because his symptoms gradually increased, he visited a hospital. Cervical radiographs taken at the initial hospital 8 days after the onset of neck pain showed a mild AAS (Figure 1A, arrow) and os odontoideum (Figure 1A, double arrows). Magnetic resonance (MR) images 19 days after the onset show a prevertebral mass at the C1–C4 level of high intensity on the T2-weighted image (Figure 1B, arrowhead). Because no apparent compression of the spinal cord was seen, he was treated with anti-inflammatory drugs. However, his neck pain and gait disturbance became aggravated. Thus, he was referred to our hospital for further examination and treatment.

On admission, he was 159 cm tall and weighed 92 kg, showing a typical obesity. He suffered from severe neck pain and stiffness. The white blood cell count was 8700, and C-reactive protein was 1.9 mg/L. He was unable to walk independently and needed a wheelchair for movement. His bladder function was disturbed, showing urinary retardation. Because he had mental retardation, it was difficult to precisely evaluate his sensory and motor loss.

Cervical radiographs obtained at admission showed severe AAS (Figure 2), indicating an atlantodental interval (ADI) of 17 mm. The anterior displacement of C1 on C2 was not reduced, even on extension of the subject's neck (Figure 2A, arrow). MR images showed the compression of the spinal cord posteriorly and anteriorly at

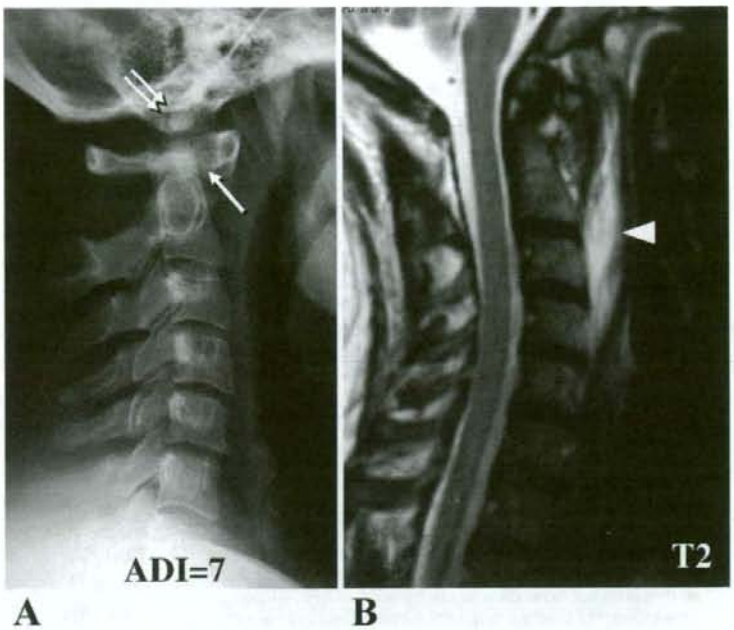

Figure 1. These 2 images of the cervical spine (**A, B**) were obtained shortly after the onset of neck pain in a 26-year-old man with Down syndrome. A plain lateral radiograph 8 days after the onset (**A**) shows a mild atlantoaxial subluxation (AAS), indicating an atlantodental interval (ADI) of 7 mm (arrow). At the craniovertebral junction, os odontoideum is present (double arrows). A midsagittal section of a T2-weighted (**B**) magnetic resonance (MR) image 19 days after the onset shows a high-intensity prevertebral mass at the C1–C4 level (arrowhead).

the C1 level (Figure 3). The T2-weighted image showed a prevertebral high-intensity area at the C2–C3 level (Figure 3C, arrowhead); the size of the high-intensity area was decreased compared with that observed at the previous hospital (Figure 1B, arrowhead). The gadolinium-enhanced image showed enhancement around the odontoid process of the axis and the anterior arch of the atlas (Figure 3B, arrow).

Treatment With Halo-Vest

We diagnosed that the patient had AAS related to the infection at the retropharyngeal space. The patient received a 6-week course of wide-spectrum antibiotics, and his upper cervical spine was stabilized with a halo-vest. After the treatment, his symptoms were gradually relieved. The cervical radiograph, 3 days after the application of the halo-vest, showed a reduction of AAS, with an ADI of 6 mm (Figure 4A, arrow).

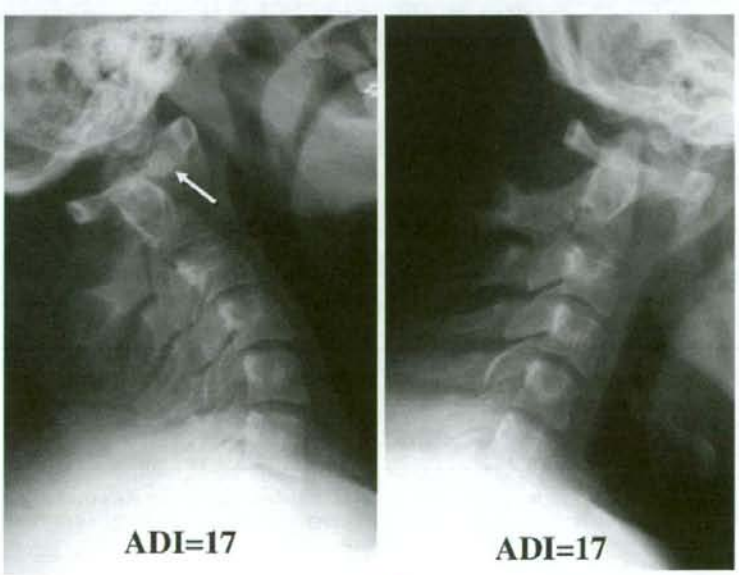

Figure 2. Extension (**A**) and flexion (**B**) cervical radiographs obtained at admission to our hospital (8 weeks after the onset of neck pain) show severe AAS, indicating an ADI of 17 mm. The anterior displacement of C1 was not reduced, even with the extension of the subject's neck (arrow).

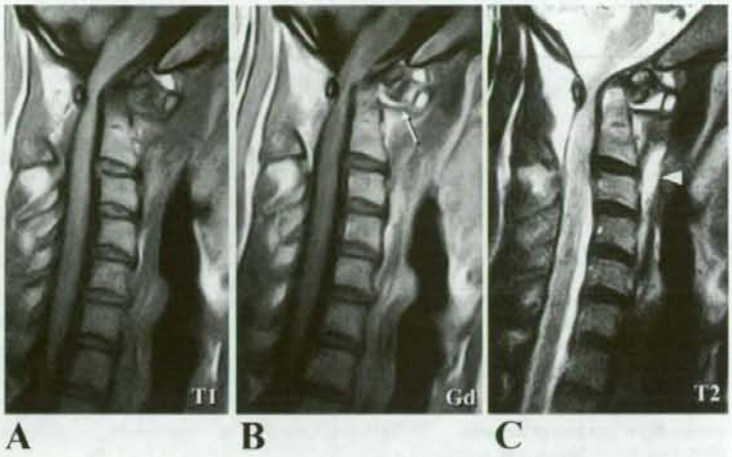

Figure 3. Midsagittal sections of T1-weighted (**A**), gadolinium (Gd)-enhanced (**B**) and T2-weighted (**C**) MR images obtained at admission to our hospital show compression of the spinal cord posteriorly and anteriorly at the C1 level. The T2-weighted image shows a prevertebral high-intensity area at the C2–C3 level (**C**, arrowhead). The Gd-enhanced image shows enhancement around the odontoid process of the axis and the anterior arch of the atlas (**B**, arrow).

After the patient was fitted with a halo-vest for 3 months, his neck pain was nearly relieved. We therefore removed the halo-vest, and applied a Philadelphia collar. Immediately after the removal of the halo-vest, however, the patient felt neck pain again. His neck pain gradually increased, and he complained of motor loss in his upper and lower extremities and urinary disturbance. One month after the removal of the halo-vest, his symptoms became prominent; he could not stand up independently and he suffered from complete urinary retention. Cervical radiographs 4 days after the removal of the halo-vest and application of the Philadelphia collar showed a recurrence of AAS, with an ADI of 17 mm (Figure 4B, double arrows).

The patient was again fitted with the halo-vest. After reapplication of the halo-vest, his symptoms were gradually relieved. Cervical radiographs 2 days after the reapplication of the halo-vest showed an incomplete reduction of AAS, with an ADI of 13 mm (Figure 4C, arrowhead).

Operation

We felt that conservative treatment with the halo-vest would be ineffective for this patient, and selected a surgical treatment. The surgical procedure we initially planned consisted of a laminotomy of the C1 posterior arch in conjunction with posterior occipitocervical fusion with instrumentation. Taking into account the MR findings that the CVJ area was enhanced with gadolinium, we thought that the risk of postoperative infection would be increased if we inserted screws into the CVJ area. Thus we selected a procedure of occipito-C3 fusion with a titanium fan-shaped rod and multistrand polyethylene cables.

During the actual surgery, a considerable reduction in the degree of AAS was obtained (Figure 5A, arrow). A strut bone was harvested from the left iliac crest and grafted between the occiput and C3 lamina.

Postoperative Course

After surgery, his symptoms of neck pain and gait and urinary disturbances were almost relieved. After surgery, the patient's upper cervical spine was stabilized with a halo-vest for 6 weeks and then with a cervical collar for a further 6 weeks. MR images 6 months after surgery showed an adequate decompression of the spinal cord at C1 (Figure 5B, arrow), though posterior indentation of

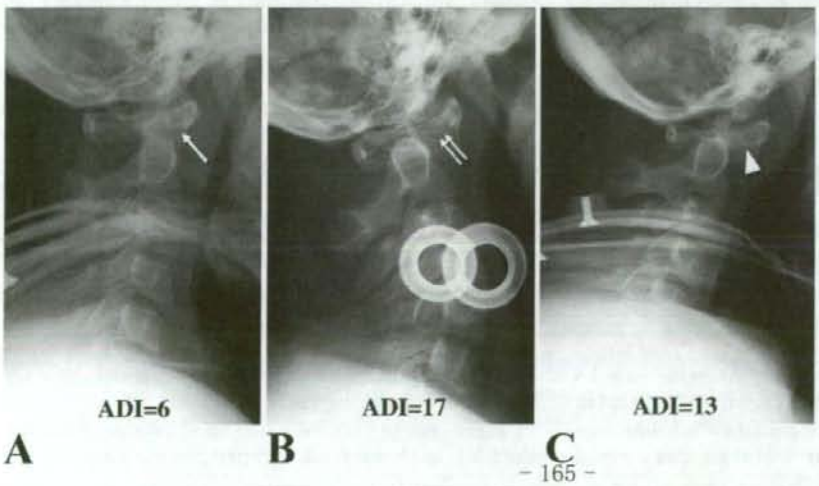

Figure 4. These cervical lateral radiographs (**A, B, C**) show the change in AAS after the patient received treatment with a halo-vest. (**A**) Three days after the application of a halo-vest (2 months and 1 week after the onset of neck pain), reduction of AAS was seen, showing an ADI of 6 mm (arrow). (**B**) Four days after the removal of the halo-vest and application of a Philadelphia collar (5 months and 3 weeks after the onset), recurrence of AAS was seen, with an ADI of 17 mm (double arrows). (**C**) Two days after the reapplication of the halo-vest (7 months after the onset), reduction of AAS occurred but was incomplete, showing an ADI of 13 mm (arrowhead).

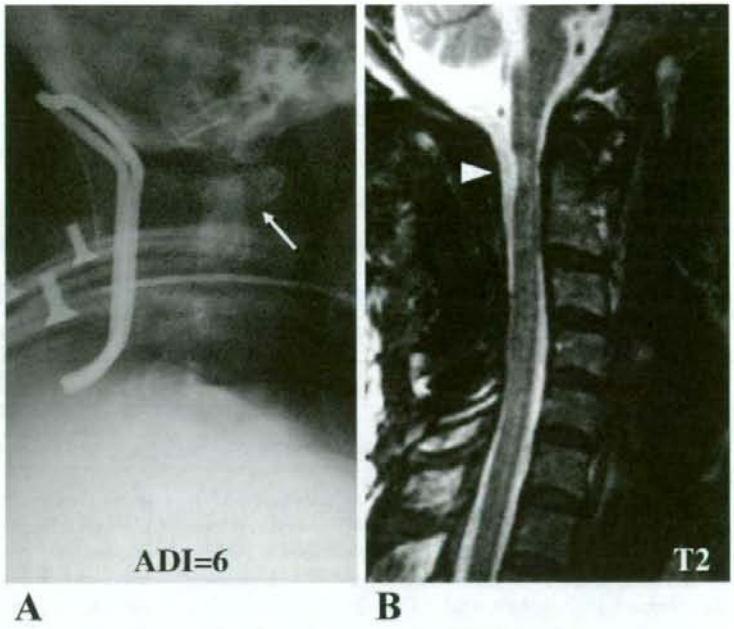

Figure 5. A lateral cervical radiograph 1 week after surgery (**A**) shows a reduction of AAS (ADI = 6 mm) (arrow) after laminectomy of the C1 posterior arch and posterior occipito-C3 fusion with titanium instrumentation and multistrand polyethylene cables. Midsagittal sections of T2-weighted (**B**) MR images 6 months after surgery show adequate decompression of the spinal cord at C1 (**B**, arrowhead). Posterior indentation of the spinal cord and a hyperintense signal within the spinal cord were seen at the C1 level.

the spinal cord and a hyper-intense signal within the spinal cord were seen at the C1 level. At a follow-up 3 years after the surgery, spinal fusion was obtained between the occiput and C3, and no symptoms of neck pain or gait and urinary disturbances were seen.

■ Discussion

Grisel syndrome has been characterized as a nontraumatic AAS associated with infection at the pharynx and its surrounding tissues.[8] Previous reports have shown that this syndrome occurs after pharyngitis, nasopharyngitis, adenotonsilitis, tonsillar abscess, parotitis, cervical abscess, and otitis media.[8–11] The developmental mechanism of Grisel syndrome has been explained as follows. First, infection around the pharynx extends to the synovial joints, joint capsule, and ligaments at the CVJ through the periodontoidal venous plexus.[8,9] Because no lymph node is present in this plexus, septic exudates may be freely transferred from the pharynx to the C1–C2 articulation. Then, the transferred infection damages the tissues at the CVJ both mechanically and chemically. The transverse ligament plays a role in preventing excessive anterior shift of the atlas on the axis. In Grisel syndrome, it is believed that hyperaemia after infection decalcifies the anterior arch of the atlas, particularly around the sites of ligamentous attachment to bone, causing the loosening of the transverse ligament.

According to previous reports, 75% to 85% of Grisel syndrome cases present in children younger than 13 years of age,[8] and this syndrome is relatively unusual in adults.[10,11] Several authors have described how ligamentous laxity and greater vascularity at the upper cervical spine in childhood contribute to the development of AAS in Grisel syndrome cases.[8]

Most patients with Grisel syndrome complain of neck stiffness and pain; however, neurologic complications are uncommon in this syndrome.[8,10] Previous reports emphasized the importance of early diagnosis and treatment with the administration of antibiotics and stabilization of the upper cervical spine with a cervical orthosis at an early stage after the onset of symptoms. In most cases, symptoms were relieved by conservative treatments, and indications of surgical treatment were limited.[8,10,11]

To the best of our knowledge, this is the first report that describes the development of Grisel syndrome in a patient with Down syndrome. In the present case, although the patient is an adult, retropharyngeal infection caused the aggravation of AAS and the development of myelopathy. For treatment, we immediately performed antibiotic therapy and immobilized his upper cervical spine with a halo-vest for 3 months, as previous reports recommended.[8,10,11] However, the conservative treatments failed to reduce the AAS, and surgical correction was required to relieve his symptoms.

In Down syndrome patients, including adults, there is an anatomic predisposition to ligamentous laxity, including the transverse ligament at the CVJ.[1] We speculate that in the present case, this laxity is one of the reasons why retropharyngeal infection caused the symptomatic AAS and why the halo-vest treatment was unsuccessful in reducing the AAS.

In the present case, the patient had os odontoideum at the CVJ. In Down syndrome patients with a congenital

skeletal anomaly such as os odontoideum, atlantoaxial instability frequently occurs, and myelopathy develops with high incidence.[5,6] We speculate that in the present case, the presence of a Down syndrome-related osseous anomaly at the CVJ decreased the efficacy of conservative treatments for repairing the damaged ligament.

Based on our limited experience, when Down syndrome patients have retropharyngeal infection, we should be aware of the possible aggravation of atlantoaxial instability and development of myelopathy, even in adults.

■ Key Points

- We report an adult case with Down syndrome, in whom infection-related atlantoaxial subluxation (Grisel syndrome) developed.
- The retropharyngeal infection aggravated the atlantoaxial subluxation, resulting in the development of myelopathy.
- Conservative treatments failed to reduce the subluxation, and surgical correction was required to relieve his symptoms.

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DIAGNOSTIC NEURORADIOLOGY

Abnormal course of the vertebral artery at the craniovertebral junction in patients with Down syndrome visualized by three-dimensional CT angiography

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Abstract
Introduction We determined the incidence of vertebral artery (VA) anomalies at the craniovertebral junction (CVJ) in patients with Down syndrome, and characterized the VA anomalies.
Methods The course of the VA in 46 consecutive patients who were due to undergo posterior arthrodesis surgery at the CVJ were evaluated by three-dimensional CT angiography (3DCTA). Included were five patients with Down syndrome who suffered from myelopathy due to atlantoaxial subluxation. All five patients with Down syndrome also had a simultaneous congenital skeletal anomaly, either os odontoideum or ossiculum terminale.
Results Of the five patients with Down syndrome, three had VA anomalies at the CVJ, two had fenestration and one had a persistent first intersegmental artery. Of the other 41 patients without Down syndrome, five had VA anomalies at the CVJ. The incidence of VA anomalies at the CVJ was much higher in patients with Down syndrome than in those without Down syndrome.
Conclusion In planning surgery in patients with Down syndrome with symptomatic atlantoaxial subluxation and a congenital skeletal anomaly at the CVJ, we should consider the possible presence of VA anomalies. Preoperative 3DCTA allows us to precisely identify an anomalous VA and evaluate the possible risk of intraoperative VA injury in advance.

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Keywords Down syndrome · Vertebral artery · Craniovertebral junction · Anomaly · Three-dimensional computed tomography angiography

Introduction

Atlantoaxial subluxation (AAS) associated with ligamentous laxity is frequently observed in patients with Down syndrome with an incidence of 12–24% [1–3], though symptomatic AAS is estimated to be present in only 1% of patients [4]. Symptoms may include cervicomedullary compression leading to the development of myelopathy. Once myelopathy occurs, decompression of the spinal cord and arthrodesis of the craniovertebral junction (CVJ) are recommended [5]. However, there have been reports describing serious intraoperative complications in surgery of the CVJ, including injuries to the vertebral artery (VA) [6, 7]. Prior to surgery at the CVJ in patients with Down syndrome, accurate detection of the course of the VA is necessary.

Previous studies using conventional catheter angiography have demonstrated anomalous courses of the VA at the CVJ [8, 9]. Two representative VA anomalies occurring in the extraosseous region include persistent first intersegmental artery and fenestration. Recently, MR angiography has provided useful information for identifying VA anomalies in the region around the cervical spine [10]. MR angiography is less invasive than catheter angiography. However, catheter angiography and MR angiography, even when combined with reconstructed CT images, cannot simultaneously show the artery and its circumferential osseous tissue, nor do they allow evaluation of the reciprocal anatomy of both tissues.

Recently, the usefulness of three-dimensional CT angiography (3DCTA) for evaluating VA anomalies at the CVJ has been reported. 3DCTA has several advantages in that it can depict the VA more precisely and can provide images reconstructed from any chosen direction, depicting the VA and circumferential osseous tissue simultaneously [11]. Since 1998, in order to avoid intraoperative VA injury, we have preoperatively evaluated VA anomalies using 3DCTA in patients due to undergo surgery at the CVJ. We have previously reported two patients with Down syndrome in whom an abnormal course of the VA at the CVJ was accurately depicted by 3DCTA [12, 13]. In the present study, we reviewed all patients who had undergone surgery for posterior arthrodesis at the CVJ in our institution. Based on the results, we evaluated the incidence of VA anomalies in patients with Down syndrome and characterized their VA anomalies.

Materials and methods

Patient population

We reviewed 46 consecutive patients who had undergone posterior arthrodesis at the CVJ at our institution between July 1998 and July 2004. Of these, 21 were male and 25 were female, with ages ranging from 5 to 81 years (mean 54.3 years). The diagnoses included AAS due to a congenital skeletal anomaly (CSA), which included os odontoideum, ossiculum terminale, hypoplastic odontoid, and/or occipitalization of the atlas (17 patients), AAS due to rheumatoid arthritis (9 patients), AAS due to dens fracture (6 patients), AAS of unknown origin (5 patients), AAS due to Down syndrome (5 patients), AAS due to cerebral palsy (3 patients), and AAS due to trauma (1 patient). All 5 patients with Down syndrome also had a CSA which included os odontoideum, ossiculum terminale and/or hypoplastic odontoid. Among the 46 patients, therefore, a total of 22 had a CSA at the CVJ and the other 24 had no osseous anomaly. The indication for operation was myelopathy (44 patients) or occipitalgia (2 patients). The operation methods were occipitocervical posterior fusion (Occ-C fusion) (31 patients) and atlantoaxial posterior fusion (C1-C2 fusion) (15 patients).

3DCTA

In all 46 patients, we performed 3DCTA before surgery and evaluated the extraosseous VA anomalies at the CVJ. From July 1998 to December 2001, we used a helical CT scanner (Somatom Plus 4; Siemens, Munich, Germany) to perform 3DCTA. Nonionic contrast medium (350 mg/ml iomeprol) was injected continuously at a rate of 3 ml/s to a volume of 2 ml/kg through a 20-gauge needle inserted into the antecubital vein. The scan parameters were as follows: delay time 15 s, shooting time 30 s, slice thickness 2 mm, and reconstruction pitch 2 mm. The source images thus obtained were transferred to a workstation (3D Virtuoso; Siemens). From January 2002 to July 2004, we used a multihelical CT scanner (LightSpeed Ultra; Yokokawa Medical Systems, Hino, Japan) and iomeprol was injected as previously described. The scan parameters were as follows: delay time 25 s, shooting time 10 s, slice thickness 1.5 mm (1.25×8 rows), and reconstruction pitch 0.875 mm. The workstation used was a Virtual Place (Office Azemoto, Tokyo, Japan).

Three-dimensional reconstructed images of the VA were obtained using the volume-rendering method. With this image, we determined the extraosseous course of the VA at the CVJ.

Statistical analysis

Statistical analysis was performed using Fisher's exact probability test. P values <0.05 were considered significant.

Results

Incidence of VA anomalies in patients with Down syndrome

VA anomalies were detected in 8 of the 46 patients reviewed. There were no complications associated with the performance of 3DCTA. No VA injuries occurred during the operation and no other complications, including neurological deficit or infection, were encountered.

In two patients with AAS due to Down syndrome, fenestration of the unilateral VA was detected. The VA was duplicated after emerging from the C2 transverse foramen. One branch entered the spinal canal between C1 and C2 and the other ran normally, passing through the C1 transverse foramen and entering the spinal canal at the cranial side of the C1 posterior arch (Fig. 1a). In one patient

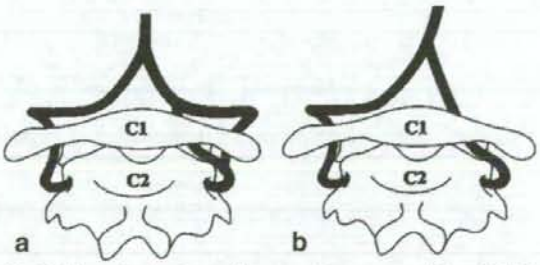

Fig. 1 Schematic drawings of the anomalous courses of the right VA at the CVJ. **a** Fenestration. **b** Persistent first intersegmental artery

with AAS due to Down syndrome and five with AAS due to CSA, a persistent first intersegmental artery (without persistence of the primary VA) was detected at the unilateral VA. The artery entered the spinal canal at the caudal portion of the C1 posterior arch after emerging from the C2 transverse foramen, without passing through the C1 transverse foramen (Fig. 1b). All eight patients with VA anomalies had congenital osseous anomalies at the CVJ, including os odontoideum, ossiculum terminale, hypoplastic odontoid, and/or occipitalization of the atlas. These eight patients underwent surgery for Occ-C fusion.

Three of the five patients with Down syndrome had VA anomalies at the CVJ. Of the other 41 patients without Down syndrome, five had VA anomalies at the CVJ. The incidence of VA anomalies at the CVJ was significantly higher in patients with Down syndrome than in patients without Down syndrome.

Characterization of VA anomalies in patients with Down syndrome

The clinical and radiographic data for the five patients with Down syndrome reviewed in the present study are summarized in Table 1. In two patients (patients 1 and 4), fenestration of the right VA was detected (Fig. 1a) [12, 13], and in one patient (patient 5), a persistent first intersegmental artery was detected at the right VA (Fig. 1b). These three patients underwent surgery for Occ-C fusion. Intraoperatively, we determined the course of the abnormal branch of the VA using Doppler ultrasonography, and carefully exposed the operative site.

Regarding the VA-related abnormal imaging findings, in the patient with the persistent first intersegmental artery (patient 5), the axial CT image at the atlas level showed that the transverse foramen was smaller on the right than on the left side (Fig. 2b). In the two patients with fenestration (patients 1 and 4) and the patient with the persistent first intersegmental artery (patient 5), axial T2-weighted MR images at the atlas level showed a circular flow-void shadow in the spinal canal (Fig. 2c).

Case presentation

Case 5

A 26-year-old man with Down syndrome presented with gait disturbance and myelopathy in association with AAS and os odontoideum (Fig. 2a). An axial CT angiography image at the atlas level showed that the transverse foramen was smaller on the right than on the left (Fig. 2b). An axial T2-weighted MR image at the atlas level showed a circular flow-void shadow in the spinal canal (Fig. 2c). The 3DCTA showed that the course of the left VA was normal, passing through the C1 transverse foramen (Fig. 3a,b). The right VA entered the spinal canal at the caudal side of C1, and did not pass through the C1 transverse foramen (Fig. 3b,c). We performed C1 laminectomy and Occ-C3 fusion with a fan-shaped rod and sublaminar wires. Autologous bone from the iliac crest was placed between the occiput and C3. Intraoperatively, the abnormal course of the right VA was identified by Doppler ultrasonography, and a surgical

Table 1 Clinical and radiographic findings for the five patients with Down syndrome who underwent surgery at the CVJ

Patient no.	Age (years)	Gender	Diagnosis	CSA	Surgery	VA anomaly	VA-related abnormal imaging findings		Reference
							MRI	CT	
1	5	M	Myelopathy, AAS, Occ-C1 instability	Hypoplastic odontoid, ossiculum terminale, bifid C1 posterior arch	Occ-C3 fusion, C1 laminectomy	Fenestration (right)	Circular flow-void shadow at C1 (right)	–	12
2	17	F	Myelopathy, AAS	Os odontoideum	C1-C2 fusion	–	–	–	–
3	15	F	Myelopathy, AAS	Os odontoideum	C1-C2 fusion	–	–	–	–
4	16	F	Myelopathy, AAS	Hypoplastic odontoid, ossiculum terminale	Occ-C2 fusion	Fenestration (right)	Circular flow-void shadow at C1 (right)	–	13
5	26	M	Myelopathy, AAS	Os odontoideum	Occ-C3 fusion, C1 laminectomy	Persistent first intersegmental artery (right)	Circular flow-void shadow at C1 (right)	Hypoplastic transverse foramen at C1 (right)	–

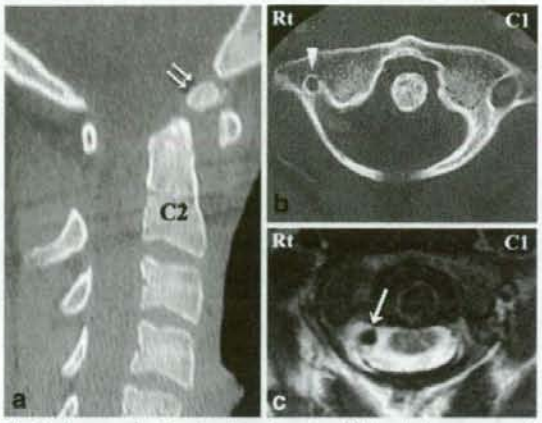

Fig. 2 Preoperative imaging in patient 5. **a** Midsagittal reconstructed image from the CT scan reveals an os odontoideum (*double arrows*). **b** Axial CTA image at the C1 level shows that the transverse foramen is smaller on the right (*arrowhead*) than on the left. **c** Axial T2-weighted MR image at the C1 level shows a circular flow-void shadow in the spinal canal (*arrow*)

approach and bone excision were undertaken with particular attention paid to avoiding injury to the anomalous VA. Following surgery, the patient's neurological deficits gradually recovered.

Discussion

Down syndrome, or trisomy 21, is the most common chromosomal disorder syndrome in humans. In general, congenital anomalies are more prevalent in children with Down syndrome than in children without Down syndrome; e.g., congenital cardiovascular anomalies [14–16] and gastrointestinal anomalies [17]. Skeletal anomalies of the CVJ are also common in patients with Down syndrome. Previous reports have shown that AAS associated with ligamentous laxity occurs in 12–24% of patients with Down syndrome [1–3]. In spite of such a high incidence of AAS, myelopathy has been reported to develop in only 1% of patients with Down syndrome [4]. Patients with Down syndrome often have osseous abnormalities at the CVJ, including os odontoideum, ossiculum terminale and/or a hypoplastic odontoid process, which has been reported at a rate of approximately 10% [3]. Previous studies have shown that the incidence of such osseous abnormalities at the CVJ in patients with Down syndrome is higher when the patients have symptomatic AAS, including myelopathy [18]. Nader-Sepahi et al. reported that 10 out of 12 patients with Down syndrome and symptomatic AAS had os odontoideum [16]. Braakhekke et al. reviewed the literature in 20 patients with Down syndrome and myelopathy, and found that 9 of the 20 patients had AAS associated with odontoid hypoplasia or os odontoideum [17].

In our institution, we performed posterior arthrodesis in five patients with Down syndrome who had AAS and myelopathy. In all five patients, os odontoideum or ossiculum terminale was present. This finding is consistent with previous reports that the presence of osseous abnormalities at the CVJ has a strong association with the development of myelopathy in patients with Down syndrome [19, 20]. Previous studies using conventional catheter angiography have shown two types of VA anomaly at the CVJ: persistent first intersegmental artery and fenestration. According to Tokuda et al., the incidences of these two types of anomaly in 300 patients who were disease-free at the CVJ were 0.67% and 1.0%, respectively [8]. In a subsequent study, Sato et al. analyzed VA angiograms of 1430 patients [9] and found that the incidences of these VA anomalies were 0.6% and 0.24%, respectively. Tokuda et al. further analyzed VA angiograms of 21 patients with osseous anomalies (occipitalization of the atlas and Klippel-Feil syndrome) at the CVJ and found

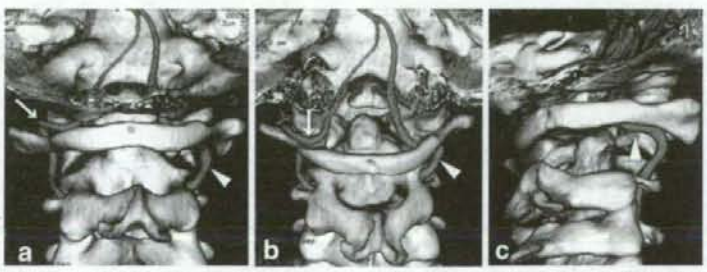

Fig. 3 Preoperative reconstructed 3DCTA images in patient 5 demonstrate a persistent first intersegmental artery for the right VA. Posterior view (**a**), posterior-superior view after deleting the occiput (**b**), and posterior-right lateral view (**c**) reconstructed 3DCTA images reveal that the right VA turns posteromedially after leaving the C2 transverse foramen and enters the spinal canal at the caudal side of the C1 posterior arch (*arrowheads*). The left VA follows the usual course, passing through the C1 transverse foramen and entering the spinal canal between the occiput and C1 (*arrow*)

persistent first intersegmental artery in 4 patients (19.0%) [8]. We have previously reported that 4 (36.4%) of 11 patients with a CSA (os odontoideum, ossiculum terminale and/or hypoplastic odontoid) who suffered from myelopathy due to AAS had extraosseous VA anomalies at the CVJ [11]. These findings have shown that the incidence of VA anomalies at the CVJ is higher in patients who also have osseous anomalies at the CVJ. Previous studies have demonstrated that the development of osseous anomalies, including occipitalization of the atlas, Klippel-Feil syndrome, os odontoideum, and ossiculum terminale, is associated with failure of resegmentation of the embryonic sclerotome [21].

In the present study, we focused on five patients with Down syndrome who suffered from myelopathy due to AAS. Interestingly, all five patients had osseous abnormalities at the CVJ (os odontoideum or ossiculum terminale). Furthermore, the incidence of VA anomalies at the CVJ was extremely high in the Down syndrome patients (three out of five) compared with that in patients without Down syndrome (5 out of 41). This finding suggests that in patients with Down syndrome who suffer from myelopathy due to AAS, there is a strong correlation between the development of a CSA at the CVJ and VA anomalies at the CVJ.

Previous studies have shown that congenital anomalies of the cardiovascular system are more prevalent in children with Down syndrome than in children without Down syndrome. Congenital heart disease has been reported to occur in approximately 40% of children with Down syndrome [14], and abnormalities of peripheral vessels are also frequent in patients with Down syndrome [15]. Regarding the VA anomaly, aberrant origins of the VA from the aortic arch have been frequently seen in patients with Down syndrome [16]. These findings collectively suggest that in patients with Down syndrome the chromosomal disorder also underlies a high incidence of developmental failure of the cardiovascular system. We speculate that, during the process of VA formation at the embryonic stage, this chromosomal disorder may strongly contribute to the failure of the development of the VA at the CVJ.

In the present study, in all three patients with Down syndrome and VA anomalies, VA-related abnormal CT and MR imaging findings were observed in the axial plane at the C1 level. In two patients with fenestration (patients 1 and 4) and one patient with persistent first intersegmental artery (patient 5), a circular flow-void shadow in the spinal canal was seen on the T2-weighted MR images. In the patient with a persistent first intersegmental artery (patient 5), hypoplasia of the unilateral C1 transverse foramen was shown on the CT image. These findings suggest that axial CT and MR images at the C1 level are useful for screening VA anomalies at the CVJ. When the presence of VA anomalies is suspected using these procedures, we should then use 3DCTA to evaluate the details of the VA course.

Compared with catheter angiography and MR angiography, 3DCTA has the following advantages: (1) accurate depiction of the VA and unrestricted image reconstruction, (2) depiction of the VA and the circumferential osseous tissue, and the ability to evaluate the reciprocal anatomy of both tissues, and (3) spatial analysis from any direction [11]. In addition, 3DCTA can be performed in a much shorter time compared with catheter angiography and MR angiography. Such advantages of 3DCTA are quite helpful for analyzing the course of the VA in patients with Down syndrome.

In conclusion, when considering surgery in patients with Down syndrome who have symptomatic AAS and osseous anomalies at the CVJ, we should consider the possibility of VA anomalies. Preoperative 3DCTA is a potential tool for precisely identifying the abnormal course of the VA. The information obtained from 3DCTA may highlight the possible risk of VA injury in advance and reduce the risk of intraoperative VA injury.

Conflict of interest statement We declare that we have no conflict of interest.

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ORIGINAL ARTICLE

Posterior instrumented fusion without neural decompression for incomplete neurological deficits following vertebral collapse in the osteoporotic thoracolumbar spine

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Abstract Previous reports have emphasized the importance of neural decompression through either an anterior or posterior approach when reconstruction surgery is performed for neurological deficits following vertebral collapse in the osteoporotic thoracolumbar spine. However, the contribution of these decompression procedures to neurological recovery has not been fully established. In the present study, we investigated 14 consecutive patients who had incomplete neurological deficits following vertebral collapse in the osteoporotic thoracolumbar spine and underwent posterior instrumented fusion without neural decompression. They were radiographically and neurologically assessed during an average follow-up period of 25 months. The mean local kyphosis angle was 14.6° at flexion and 4.1° at extension preoperatively, indicating marked instability at the collapsed vertebrae. The mean spinal canal occupation by bone fragments was 21%. After surgery, solid bony fusion was obtained in all patients. The mean local kyphosis angle became 5.8° immediately after surgery and 9.9° at the final follow-up. There was no implant dislodgement, and no additional surgery was required. In all patients, back pain was relieved, and neurological improvement was obtained by at least one modified Frankel grade. The present series demonstrate that the posterior instrumented fusion without neural decompression for incomplete neurological deficits following vertebral collapse in the osteoporotic thoracolumbar spine can provide neurological improvement and relief of back pain without major complications. We suggest that neural decompression is not essential for the treatment of neurological impairment due to osteoporotic vertebral collapse with dynamic mobility.

Keywords Neurological deficit · Osteoporosis · Vertebral collapse · Thoracolumbar spine · Posterior fusion

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Introduction

Previous reports have shown that incomplete neurological deficits following vertebral collapse in the osteoporotic thoracolumbar spine are caused by neural compression due to retropulsed bone fragments in the spinal canal, progression of kyphosis, and instability at the fracture site [1, 4, 7, 13, 17, 18]. Of the several types of surgical procedures reported for the treatment of this condition, most have emphasized the importance of decompressing the spinal cord and/or cauda equina through either an anterior or posterior approach [4, 7, 8, 13–16, 19]. However, some authors have reported that conservative treatment provides reliable neurological improvement, even without neural decompression [2, 11]. Furthermore, several studies have shown that remodeling of fractured vertebrae progresses toward recovering the normal structure of the spinal canal [5, 6]. Thus, contribution of those neural decompression procedures to neurological recovery has not been fully established.

We hypothesized that instability at the fracture site rather than neural compression is the major factor causing neurological disorders in patients with osteoporotic thoracolumbar vertebral collapse, and consequently have

performed posterior instrumented fusion without neural decompression for this condition. In the present study, we investigate the clinical outcomes of this procedure.

Materials and methods

Patient population

From August 2001 to December 2006, 14 consecutive patients with delayed neurological deficits following vertebral collapse in the osteoporotic thoracolumbar spine underwent posterior instrumented fusion without neural decompression at our institute (Table 1). The patients included ten females and four males (average age at surgery, 73.2 years; range, 46–86 years). Their average follow-up period was 25 months (range, 12–50 months). The mean duration from the onset of symptoms to surgery was 3.8 months (range, 1–12 months). Causes of osteoporosis were classified as senility-related in nine patients, rheumatoid arthritis in four, and alcoholism in one. The affected level was T8 in two patients, T10 in one, T11 in three, T12 in five, and L1 in three. Eight patients had a total of 16 old thoracolumbar compression fractures at other levels: 4 in adjacent vertebrae, 6 in vertebrae two levels above or below the affected level, and 6 in vertebrae three or more levels above or below the affected level. Their neurological functions were assessed with a modified Frankel grading system (Table 2) [3]. Five patients had deficits of grade C, six had grade D1 deficits, two grade D2 deficits, and one grade D3 deficits. Six patients had neurogenic bladder dysfunction.

Surgical techniques

In our surgical method, no procedure for decompressing the spinal cord and/or cauda equina was performed, and all patients underwent in situ posterior fusion. At surgery, the patients were moved into a prone position on the operating table with the hip joint flexed in order to reduce the thoracolumbar kyphosis. Intraoperatively, we did not attempt to correct either kyphosis or vertebral height by applying force to the implant or manual corrective adjustment. Posterior and posterolateral fusion was performed using an autologous iliac crest bone graft and a pedicle screw and rod system. We principally used pedicle screws for anchors of the instrumented fusion, and augmentation with sublaminar cables and/or hooks at multi-levels was added to prevent the pullout of the screws (Table 1). The spine was fused from three levels above to two levels below the collapsed vertebra. In eight patients, a longer segment of the spine was fused, because of the presence of concomitant old vertebral compression fractures (Table 1). The average number of fusion levels was 6.2 (range 4–9). In five patients who had large bone defect in the fractured vertebral body, transpedicular impaction of hydroxyapatite block was added to fill the defect. However, we did not perform the correction of kyphosis at this impaction. Patients were allowed out of bed with spine protected by a plastic orthosis at 4 or 5 days after the operation. They

Table 1 Data of surgeries for 14 patients

Case no.	Age (years)/ gender	Cause of osteoporosis	Affected level	Levels of old VB fx	Fusion levels	Anchors of posterior instrumented fusion		
						Pedicle screw	Sublaminar wiring	Hook
1	80/F	Senility	T12	T11, L2	T7–L3	L3	T8, T10, L2	T7, T9, L1, L3
2	67/F	Senility	T10	L1, L3, L4	T7–L3	T11, L2, L3	–	T7, T8, T9
3	74/F	Senility	T11	–	T7–L3	T10, L2, L3	–	T7, T8, L3
4	46/F	RA	T8	T6, T7	T4–T11	T4, T5, T6, T9, T10, T11	T4, T5, T6, T9, T10, T11	–
5	79/F	Senility	T11	L4	T9–L1	T9, T10, T12, L1	–	L1
6	83/F	Senility	T12	–	T9–L2	T9, T10, T11, T12, L1, L2	T9, T10, T1, T12, L1, L2	–
7	66/F	RA	L1	T12, L3	T10–L4	T10, T11, T12, L2, L3, L4	T10, T11, T12, L2, L3, L4	–
8	71/M	Senility	L1	–	T10–L2	T10, T11, T12, L2	T10, T11	L2
9	72/M	Alcoholism	T11	T7, T9, L1	T7–L2	T7, T8, T9, T10, T12, L1, L2	T7, T12, L1	–
10	72/F	RA	T12	–	T9–L3	T9, T10, T11, L1, L2, L3	T9, T10, L1, L2, L3	–
11	74/F	RA	T8	T7, T11	T4–T12	T4, T5, T6, T7, T9, T10, T1, T12	T4, T5, T6, T10, T11, T12	–
12	72/M	Senility	T12	–	T9–L2	T9, T10, T11, L2, L3	T9, T10, L2, L3	–
13	81/F	Senility	T12	T10	T9–L2	T9, T10, T11, L2, L3	T9, T10, L2, L3	–
14	86/M	Senility	L1	–	T10–L3	T10, T11, T12, L2, L3	T10, T11, L2, L3	–

VB fx vertebral fracture, *RA* rheumatoid arthritis

Table 2 The modified Frankel grading system

Grade	Neurological status
A	Complete motor loss and sensory loss
B	Preserved sensation only, voluntary motor function absent
C	Preserved motor less than fair grade (nonfunctional for any useful purpose)
D1	Preserved motor at lowest functional grade (3+/5+) and/or with bowel or bladder paralysis with normal or reduced voluntary motor function
D2	Preserved motor at midfunctional grade (3+ to 4+/5+) and/or with neurogenic bowel or bladder dysfunction
D3	Preserved motor at high-functional grade (4+ to 5+) and normal voluntary bowel or bladder function
E	Complete motor loss and sensory function normal (may still have abnormal reflexes)

remained in the plastic orthosis for 3 months, and then changed to a canvas brace for an additional 3 months.

Radiographic assessment

In preoperative radiographs, the local kyphosis angle was measured as the angle between the lower endplate of the uninvolved vertebra above the fractured level, and the upper endplate of the uninvolved vertebra below the fractured level (Fig. 1). The sagittal Cobb angle was also measured as the angle between the upper endplate of the uppermost vertebra and the lower endplate of the lowest vertebra at the instrumented fusion levels. Eleven patients underwent myelography preoperatively, and blockade of contrast medium was evaluated. In ten patients, the spinal canal occupation due to retropulsed bone fragments was measured on CT images before surgery and 6 months after surgery. Bone union of the collapsed vertebra and the instrumented fusion levels was assessed to be successful, when there was no change of the local kyphosis angle and the sagittal Cobb angle on flexion and extension radiographs. Postoperative subsequent vertebral fractures and complications related to the surgical instrumentation were also analyzed.

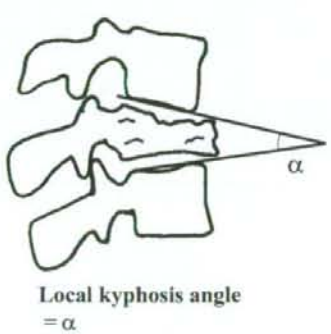

Fig. 1 Schematic diagrams of radiographic measurements

Clinical assessment

Back pain was classified into four grades: none, mild, moderate, and severe. Mild pain was defined as intermittent pain during motion; moderate pain was defined as pain preventing the patient from sitting in a chair, but without pain while resting in bed; and severe pain was defined as pain even while resting in bed. Clinical outcomes for back pain were assessed using this classification. In addition, neurological function was evaluated using a modified Frankel grading system (Table 2) [3].

Results

Before surgery, the mean local kyphosis angle was 14.6° (range, 0–40°) on flexion radiographs and 4.1° (range, −18–27°) on extension radiographs (Table 3). Thus, the mean change in the local kyphosis angle in flexion and extension was 10.6° (range, 7–18°), demonstrating marked instability at the collapsed vertebrae. Intraoperatively, there was no finding of deficiency and attenuation in the supra- and inter-spinous ligaments in 14 patients, indicating little participation of the posterior elements of spinal column to the instability.

Myelogram showed no obstruction of the contrast medium in all 11 patients examined. Before surgery, the mean spinal canal occupation by retropulsed bone fragments was 21.1% (range, 0–37.4%). Among ten patients examined, the spinal canal compromise was 0% before surgery in two patients. In the remaining eight patients, remodeling of the spinal canal was observed (Fig. 4e, f, g). The average spinal canal compromise was decreased from 26.3% before surgery to 18.1% at 6 months after surgery. Anterior spinal canal encroachment due to factors other than bone fragments was shown on MRI; granulation tissue was found in the vertebral body of one patient with pseudoarthrosis and hematoma in another patient, both conditions resolved spontaneously after surgery.

Table 3 Alteration of the local kyphosis angle at the affected vertebra

Case no.	Local kyphosis angle (°)				
	Before surgery		Immediately after surgery	Six months after surgery	Final follow-up
	Flexion	Extension			
1	21	9	10	18	18
2	0	−18	−10	−4	−3
3	19	11	8	15	16
4	25	19	25	28	28
5	6	−1	0	10	10
6	18	5	10	13	14
7	5	−6	−8	−5	−5
8	11	3	0	4	4
9	0	−7	−7	−5	−5
10	28	18	18	18	17
11	40	27	29	30	30
12	11	3	5	5	6
13	15	2	6	6	6
14	9	−8	−5	0	2
Mean ± SD	14.6 ± 11.2	4.1 ± 12.1	5.8 ± 12.0	9.5 ± 11.5	9.9 ± 11.3

The mean local kyphotic angle in flexion before surgery (14.6°; Table 3) was significantly reduced to 5.8° immediately after surgery, 9.5° at 6 months after surgery, and 9.9° at the final follow-up (Table 3). The mean correction of the local kyphosis was 8.9° immediately after surgery and 4.7° at the final follow-up; thus the mean loss of correction was 4.1°. There was no implant dislodgement, and no additional surgery was required. Solid union of both parts of the collapsed vertebral body as well as posterior and posterolateral spine fusion was successfully achieved in all patients.

In seven patients, subsequent vertebral compression fractures developed after surgery within the fusion level and/or at adjacent or nearby vertebrae above or below the fusion level (Table 4). All the subsequent fractures were well managed conservatively, and no patients complained residual back pain at the final follow-up. Increase of local kyphotic angle after the onset of each subsequent fracture was within 5° at the final follow-up (Table 4), indicating no development of the junctional kyphosis [9].

All patients had back pain before surgery. The pain grades in 12 patients were moderate or severe. In all 14 patients, back pain was relieved after surgery. At the latest follow-up assessment, 11 of 14 patients reported complete relief of pain (Fig. 2). In all patients, neurological function improved by at least one modified Frankel grade (Fig. 3).

Table 4 Data of seven patients with the subsequent fracture after surgery

Case no.	Fusion levels	Levels of subsequent fx	Surgery to subsequent fx (mo)	Local kyphosis angle at subsequent fx (°)	
				Immediately after surgery	Final follow-up
1	T7–L3	T8	1	1	5
		T10	6	3	3
		L1	12	16	20
2	T7–L3	T7	4	6	11
		T5	36	2	7
3	T7–L3	T6	3	7	9
		L4	36	−15	−12
8	T10–L2	L4	3	−16	−13
		L2	6	−3	1
10	T9–L3	T9	1	4	7
		L5	12	−34	−31
11	T4–T12	T4	1.5	14	15
12	T9–L2	T8	17	2	7

Subsequent fx = subsequent fracture