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Surgical treatment for cervical spondylotic myelopathy associated with athetoid cerebral palsy

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Abstract There have been several reports on surgical interventions in patients with adult cervical spondylotic myelopathy associated with athetoid cerebral palsy; however, the long-term effectiveness of these interventions has not been demonstrated. We have performed surgical treatments — posterior fusion with wave-shaped rods and anterior interbody fusion with internal fixators — in 20 patients. The present study included 17 of these patients, 16 men and 1 woman, and their mean follow-up period was 8.6 years (range, 5–15.5 years). One year after surgery, walking ability was improved in 14 patients. Pain in the upper extremities and muscle weakness of the deltoid were alleviated in all patients. One patient showed recurrence of myelopathy after 8.5 years' follow-up. Our surgical technique is effective in patients with cervical spondylotic myelopathy secondary to athetoid cerebral palsy, even in those with severe involuntary movements. Postoperative rigid external fixations are not required.

Key words Cervical spondylotic myelopathy · Athetoid cerebral palsy · Surgical treatment

Introduction

Cervical spondylotic myelopathy, which is frequently associated with athetoid cerebral palsy, is a serious secondary disability for the people who already have this congenital handicap. It causes considerable decline in their daily life activities.[4] Among treatments for this disease, conservative therapy such as cervical orthosis and/or medication is not appropriate, due to the involuntary movements. Although several surgical procedures have been described in the literature, none of them yielded satisfying long-term results.[7,17] The disease is frequently associated with nerve root neuropathy, which causes pain in the upper extremities and decline of muscular strength.[14,18] We suggest the hypothesis that severe instability of the spine induced by involuntary movements, malalignment of the cervical spine due to the long-lasting strain of cervical muscle imbalance, and the compression against neural elements by spurs from cervical spondylotic changes are the main pathologic factors leading to the serious disability. In 1983,[1] we developed a surgical technique, anterior posterior fusion using wave-shaped rods, that can eliminate these three causes, and the procedure has since been applied to clinical practice.[20] In this study, we report the patients' clinical symptoms, the radiographic findings, and the surgical results of the technique, which is described as well.

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Patients and methods

Patients

From June 1983 to November 1993, at our hospital, we performed surgical operations in 20 patients suffering from cervical spondylotic myelopathy secondary to athetoid cerebral palsy. We excluded 3 patients from the study because full 3-year follow-up after surgery was not possible. One of them was transferred to a care center far from our hospital, and the other 2 could not undergo follow-up because of walking disability. Seventeen patients had been followed for more than 5 years, and the mean follow-up period was 8.6 years (range, 5 to 15.5 years). There were 16 men and 1 woman. Average age at the time of surgery was 44.5 years (range, 19 to 62 years). Sixteen patients could walk without aid (1 was walking with a cane) before they experienced cervical spondylotic myelopathy symptoms. However, on admission, 5 patients could not walk, 8 could walk with aid, and only 4 could walk without aid. Eight patients had pain in the upper

Fig. 1. Radiographic measurements. The Degree of motion at the intervertebral disc adjacent to the fusion area = $(A + B)°$, where A is the flexion angle and B is the extension angle. The Atlanto-dental interval in the flexed position = C (mm). Retrolisthesis in the extended position = D (mm)

extremities. According to the Denis Pain Scale,[5] these patients were classified as: grade three (1 patient), grade four (6 patients), and grade five (1 patient). Manual muscle testing (MMT) revealed deltoid muscle weakness in 8 patients, of whom 7 patients were classified as grade 2 and one patient as grade 4. The average duration of deterioration, beginning with the appearance of numbness in the extremities, and continuing with the decline of muscular or walking abilities that led to surgery was 1 year and 9 months (range, 1 month to 10 years).

Radiographic findings

Preoperative cervical roentgenograms for all 17 patients showed cervical spondylotic changes, malalignment, and/or some degree of instability (anterior or posterior spondylolisthesis). No subluxation between C1 and C2 was seen. The average sagittal diameter at the C5 level of the spinal canal was 13.8 mm (range, 11.5 to 16.0 mm, after adjustment for magnification). Eleven patients had spinal stenosis with the sagittal diameter of the spinal canal less than 14 mm. We performed myelography and computed tomography after myelography for all patients to identify the exact sites of neural compression. The results showed that 2 patients had a single level intervertebral lesion between C3/4, while the remaining 15 patients had multiple intervertebral lesions. Of these 15 patients, 8 had an intervertebral lesion at C3–6, 2 patients had this lesion at C3–7, 2 had this lesion at C4–6, 2 had this lesion at C4–7, and 1 patient had this lesion at C2–5.

Operative technique

Preoperative external support with a halo vest was applied in 2 patients, skull traction was applied in 2 other patients, and in the remaining 13 patients, who were treated after 1988, a cervical soft collar was used. The purposes of the surgical operation were to decompress the spinal cord and nerve roots, to correct malalignment of the cervical spine, and to gain stability.

Table 1. Method of clinical evaluation

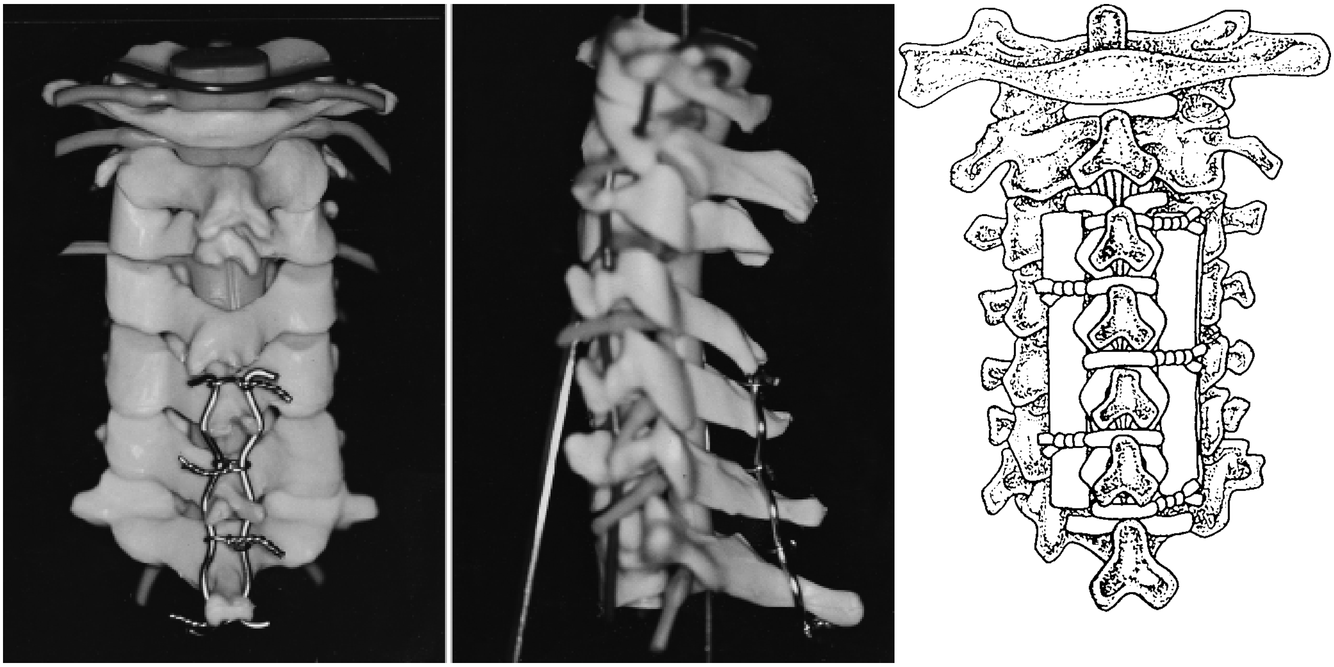
Walking ability		
	Grade 1	Able to walk without aid
	Grade 2	Able to walk with aid
	Grade 3	Need for wheel chair
Pain scale (Denis[5])		
	Grade 1	No pain
	Grade 2	Occasional minimal pain with no need of medication
	Grade 3	Moderate pain with occasional medication, but no interruption of work or significant change in daily life activities
	Grade 4	Moderate to severe pain with frequent medication and occasional absence from work or significant change in daily life activities
	Grade 5	Constant or severe incapacitating pain with chronic medication

With the patient under general anesthesia, a posterior median incision was made, with the patient in the prone position, which exposed the neural arches and spinous processes. Using preoperative myelograms, the proper cervical alignment was determined. Wave-shaped rods, a pair of 1.8-mm diameter rods, were applied to maintain the alignment, thus altering the interspinous intervals. After confirmation of stability, the neural arches were decorticated, and posterior fusion, using unicortical and cancellous iliac bone, was performed (Fig. 2) Then we changed the patient's position to supine, and performed anterior cervical decompression by removing the bony rim and osteophytes after complete discectomy. We, basically, did not perform subtotal spondylectomy. Iliac bone was grafted to all intervertebral spaces in accordance with the method of Smith and Robinson.[24] Then, in 8 patients, we fixed the bone graft with a plate and screw system (Fig. 3). In 9 patients whose vertebral bodies collapsed, we did not use a plate because it was difficult to insert a screw through a hole in the plate. In these patients, we inserted screws into the vertebral body without a plate and then the head of each screw was fixed to the head of each other screw with 0.8-mm or 1.0-mm diameter wires (Fig. 4). All screws were inserted with unicortical purchase. After checking the positioning of the screws radiographically, we closed the incision with a drain tube in place.

The 4 patients who were treated before 1988, and for whom halo vest and skeletal traction had been applied, were allowed to sit 2 weeks after the surgery. The 13 patients in whom a cervical soft collar was used were allowed to sit 3 to 7 days after the surgery.

Clinical evaluation

Walking ability was classified into three grades (Table 1); grade 1, able to ambulate without help; grade 2, able



A,B C

Fig. 2A–C. Method of posterior fusion with wave-shaped rod (Wavy rod, Best Medical, Tokyo). **A** Posterior view; **B** lateral view; **C** schematic drawing of the postoperative view. The spinous process was held with two wave-shaped rods (Wavy rod) that were made in order to correct the spinal malalignment, and then two rods were fastened with wires in the interspinous spaces. Finally, the iliac bone was grafted

to ambulate with a cane and/or help; grade 3, unable to ambulate and moving in a wheel chair. Pain in the upper extremities was evaluated by classifying it into five grades according to Denis' Pain Scale.[5] Deltoid muscle strength was evaluated, using the MMT classification, and classified into six grades. Evaluations were done at the preoperative visit, 1 year after the surgery, and at final follow-up. At final follow-up, results were classified as: excellent, improvement of more than two grades; good, improvement of one grade; and poor, no improvement, or deterioration.

Postoperative roentgenograms were evaluated from the following four aspects:
(1) Motion in operative sites with functional radiographs
(2) Angular motion and spondylolisthesis at levels adjacent to the fixed segments (Fig. 1A,B,D)
(3) Atlanto-axial subluxation (Fig. 1C)
(4) Erosion of spinous processes (Fig. 5B).

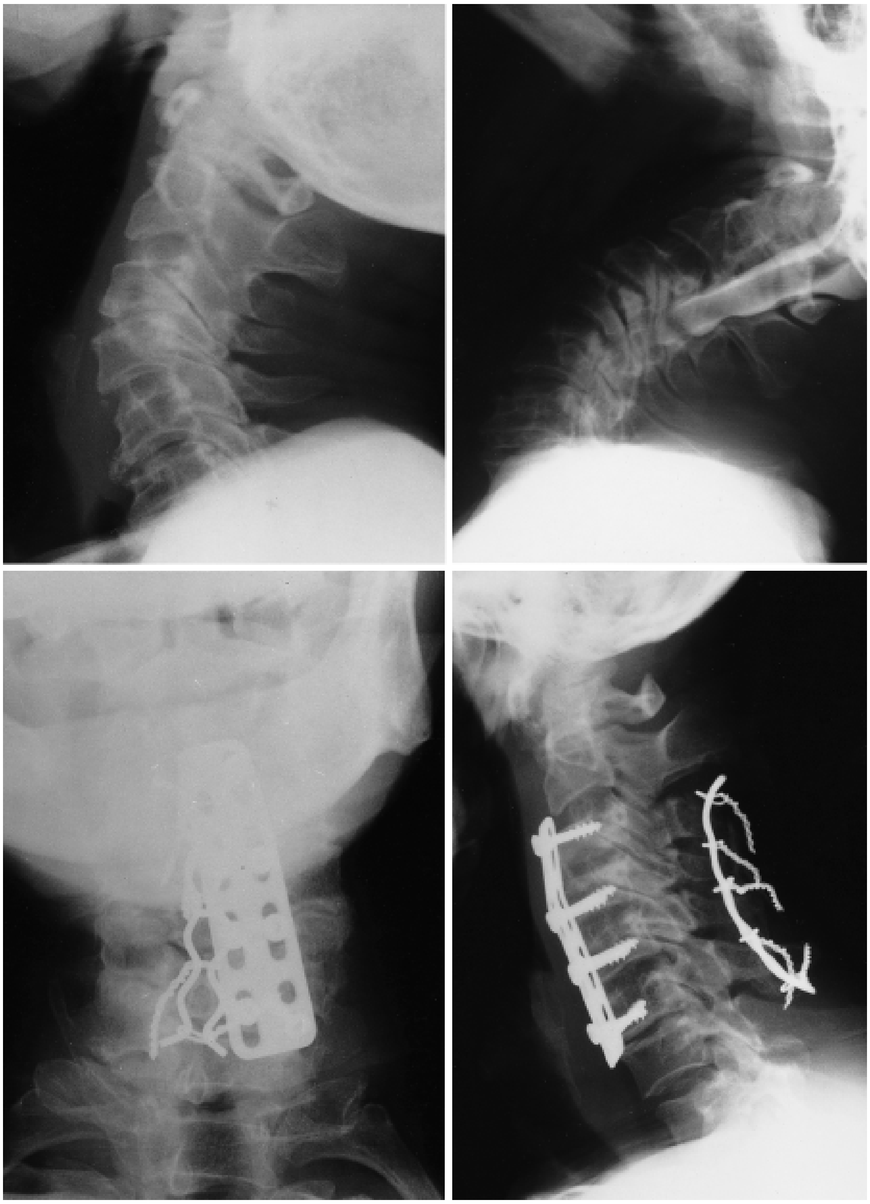
Results

Clinical results

Regarding walking ability, 4 patients were classified as grade 1, 8 as grade 2, and 5 as grade 3 at the preoperative evaluation. The 4 grade 1 patients remained in the same grade until the final follow-up. Of the remaining 13 patients, 3 were classified as excellent (improved from grade 3 to grade 1), 7 as good (improved from grade 2 to grade 1 or from grade 3 to grade 2), and 3 as poor (cases 3, 12, and 15) at the final follow-up. At final follow-up, walking ability had declined in 3 patients (cases 2, 10, and 11) (Tables 2, 3). The causes of the decline in walking ability were considered to be as follows: a lumbar disorder had developed in 1 patient (case 2), retrolisthesis at the level adjacent to the fusion site had advanced in the second patient (case 10), and long-term bed rest due to physical disorder had led to deterioration of general muscle strength in the third patient (case 11).

Regarding pain in the upper extremities, one patient (case 7) was in grade 3, six patients (cases 2, 3, 12, 15, 16, and 17) in grade 4, and one (case 11) in grade 5, preoperatively. The treatment results were excellent in five patients (cases 2, 11, 15, 16, and 17) good in three (cases 3, 7, 12), and there was no patient with poor results at 1 year follow-up. But at the final evaluation, there were three patients in whom deterioration was observed (cases 11, 12, and 14) (Tables 2, 3).

Concerning the decline of deltoid muscular strength, seven patients (cases 1, 2, 4, 5, 9, 10, and 13) were classified preoperatively as grade 2 by MMT, and one



A B C D

Fig. 3A–D. Roentgenograms of case 11. **A** Lateral view and **B** myelogram showing instability, malalignment, and bony spurs that compressed the dural tube between C3 and C6. **C** Anteroposterior and **D** lateral radiographs made 8 years after the anterior posterior cervical fusion, revealing decreased retrolisthesis by the spreading of the interspinous space between the third and fourth cervical levels; resection of posterior bony spurs; complete bony union; and correction of malalignment

patient (case 17) as grade 4. One year after the surgery, in regard to improvement in deltoid muscular strength, the results were classified as excellent in six patients (cases 1, 2, 4, 9, 10, and 13), good in one patient (case 5), and no improvement was observed in one patient (case 17) whose grade (grade 4) did not change. Results at the final evaluation were almost the same as those 1 year after the surgery (Tables 2, 3).

Complications

Hypertonia of athetoid movements due to pain stimuli was observed in all patients until 2 weeks after the surgery. Some patients complained of difficulties in talking and/or swallowing because movement of the cervical spine was limited by the fusion; however, they did not have such complaints after 2 weeks postoperatively. Involuntary movements decreased in general; however, the degree of decrease varied depending on the patient's preoperative movements and the fusion levels. No serious complications were observed.

Radiographic results

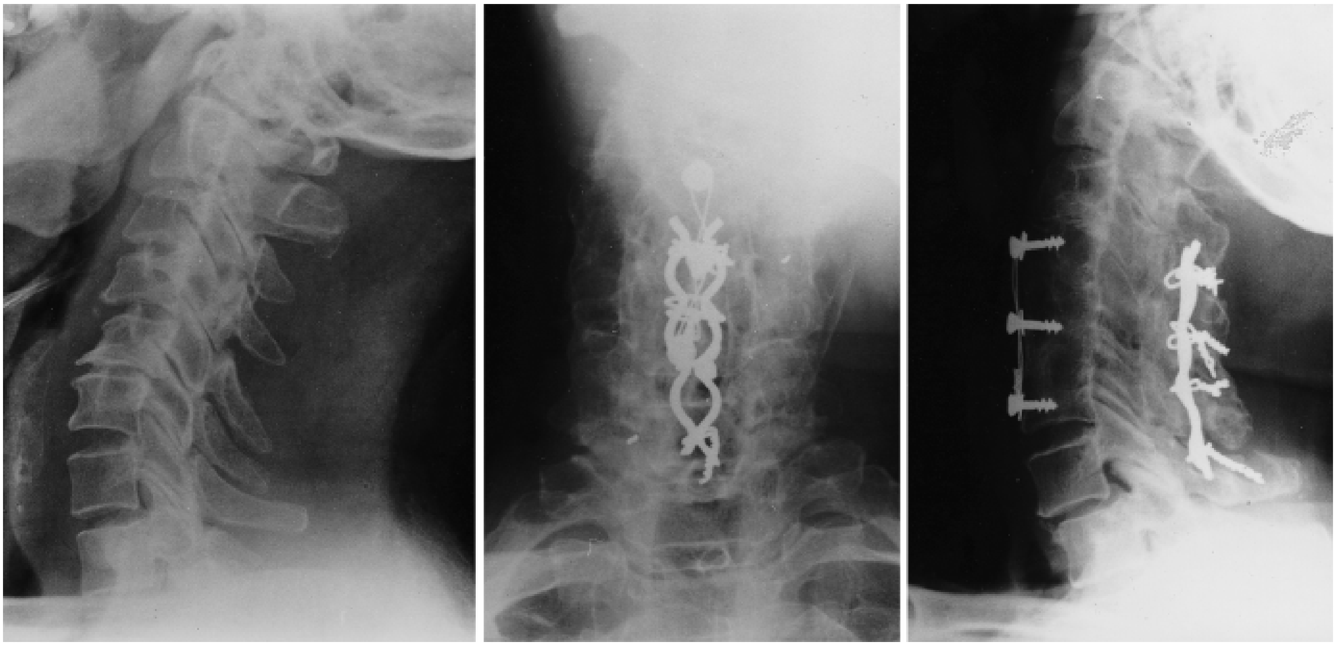
We confirmed bone consolidation in all patients within 6 months after the surgery. A screw which was inserted into the vertebral body was broken in one patient (case 12), but reoperation was not required because the screw

Table 2. Clinical and radiographic data before operation

Case no.	Age (years) at op.	Sex	Period from onset to admission (months)	Symptom	Walking ability before onset (grade)	Ability of walking at admission (grade)	Brachialgia (pain scale; grade)	Weakness of the deltoid muscle (MMT)	Protruding osseous spur on myelogram (cervical level)	Sagittal diameter of the spinal canal at C5 (mm)
1	44	M	5	Myeloradiculopathy	1	2	1	2	4/5/6	15
2	48	F	3	Myeloradiculopathy	1	3	4	2	3/4/5/6	13
3	53	M	3	Myeloradiculopathy	1	2	4	5	3/4/5/6	13
4	52	M	14	Myeloradiculopathy	1	1	1	2	4/5/6	14.5
5	39	M	2	Myelopathy	1	1	1	2	2/3/4/5	15
6	37	M	3	Myeloradiculopathy	1	2	1	5	3/4/5/6/7	13
7	49	M	2	Myelopathy	2	3	3	5	3/4/5/6	13.5
8	45	M	1	Myeloradiculopathy	1	3	1	5	3/4/5/6/7	13
9	31	M	24	Myeloradiculopathy	1	2	1	2	3/4/5/6	15
10	50	M	15	Myeloradiculopathy	1	2	1	2	3/4	15
11	62	M	18	Myeloradiculopathy	1	3	5	5	3/4/5/6	14
12	47	M	120	Myeloradiculopathy	1	2	4	5	3/4/5/6	13.5
13	41	M	31	Myelopathy	1	2	1	2	3/4/5/6	14
14	42	M	3	Myeloradiculopathy	1	2	1	5	3/4/5/6	12
15	47	M	96	Myeloradiculopathy	3	3	4	5	4/5, 6/7	11.5
16	19	M	14	Myeloradiculopathy	1	1	4	5	3/4	13.5
17	36	M	12	Myeloradiculopathy	1	1	4	4	4/5/6/7	16

op., Operation; MMT, manual muscle testing had not backed out. Slight screw backouts were seen in two patients, although these backouts stopped after bone consolidation, and the screws remained in the same positions (cases 11 and 14).

The mean motion angle at the upper level adjacent to the fixed segments was increased from a preoperative value of 4.5° (range, 0°–14.0°) to 7.2° (range, 0°–10.0°) after the operation; however, there was no significant difference between the pre- and postoperatively observed angles. Three-mm spondylolisthesis of a vertebral body posteriorly was seen in one patient (case 17). At the lower level adjacent to the fusion site, the



A,B C

Fig. 4A–C. Roentgenograms of case 1. **A** Lateral radiograph before surgery, showing malalignment, deformity of vertebral body, and posterior spurs between C4 and C6. **B** Anteroposterior and **C** lateral radiographs made 15 years after the anterior and posterior cervical fusion, revealing correction of malalignment and complete bony fusion between C4 and C6, and spontaneous posterior fusion at the third and fourth cervical levels

Table 3. Clinical and radiographic data after operation

Case no.	Period of follow-up (Years + months)	Surgical treatment: Anterior fusion level	Surgical treatment: Instrument	Surgical treatment: Posterior fusion level	Surgical treatment: Instrument	Walking ability 1 year after op. (grade)	Ability of walking at follow-up (grade)	Brachialgia (pain scale) 1 year after op. (grade)	Brachialgia (pain scale) at follow-up (grade)
1	15 + 6	C4–6	S&W	C4–6	WSR	1	1	1	1
2	13 + 1	C3–6	S&W	C3–6	WSR	1	2	1	1
3	8 + 7	C3–6	Plate	C3–6	WSR	2	2	3	3
4	12 + 2	C4–6	S&W	C4–6	WSR	1	1	1	1
5	10 + 6	C2–5	S&W	C2–5	WSR	1	1	1	1
6	10 + 0	C3–7	Plate	C3–7	WSR	1	1	1	1
7	5 + 10	C3–6	Plate	C3–6	WSR	2	2	2	2
8	9 + 9	C3–7	S&W	C3–7	WSR	1	1	1	1
9	9 + 7	C3–6	—	C3–6	WSR	1	1	1	1
10	8 + 6	C3/4	Plate	C3/4	WSR	1	2	1	1
11	7 + 8	C3–6	Plate	C3–6	WSR	1	3	3	4
12	7 + 6	C3–6	Plate	C3–6	WSR	2	2	3	4
13	6 + 9	C3–6	Plate	C3–6	WSR	1	1	1	1
14	5 + 10	C3–6	Plate	C3–6	WSR	1	1	1	3
15	5 + 3	C4/5,6/7	S&W	C4–7	WSR	3	3	1	1
16	5 + 0	C3/4	S&W	C3/4	WSR	1	1	1	1
17	5 + 0	C4–7	S&W	C4–7	WSR	1	1	1	1

S&W, Screw and wire; WSR, wave-shaped rod mean motion angle was 9.1° (range, 0°–20.0°) preoperatively, and 7.4° (range, 0°–20.0°) at the final evaluation. There was no significant difference between these angles. Spondylolisthesis of a vertebral body posteriorly was observed in 1 patient (case 10). Intervertebral instability at the level adjacent to the fusion site was noted only in these two patients with spondylolisthesis (cases 10 and 17). On the other hand, stabilization at the level adjacent to the fusion site was observed in 6 patients (cases 1, 2, 4, 7, 9, and 17) cranially, and in 7 patients (cases 1, 2, 5, 7, 11, 12, and 15) caudally. Bone consolidation was found in 4 patients (cases 1, 2, 4, and 12) (Fig. 6) at the upper level adjacent to the fusion site and in 3 patients (cases 4, 11, and 15) at the lower level adjacent to the fusion site. Subluxation between C1 and C2 was not detected preoperatively, but in 4 patients it was observed at the final evaluation (cases 4, 5, 11, and 12). However, these subluxations were slight, and the average atlanto-dental interval in the flexed position was 6.0 mm (range, 5.0–

A B

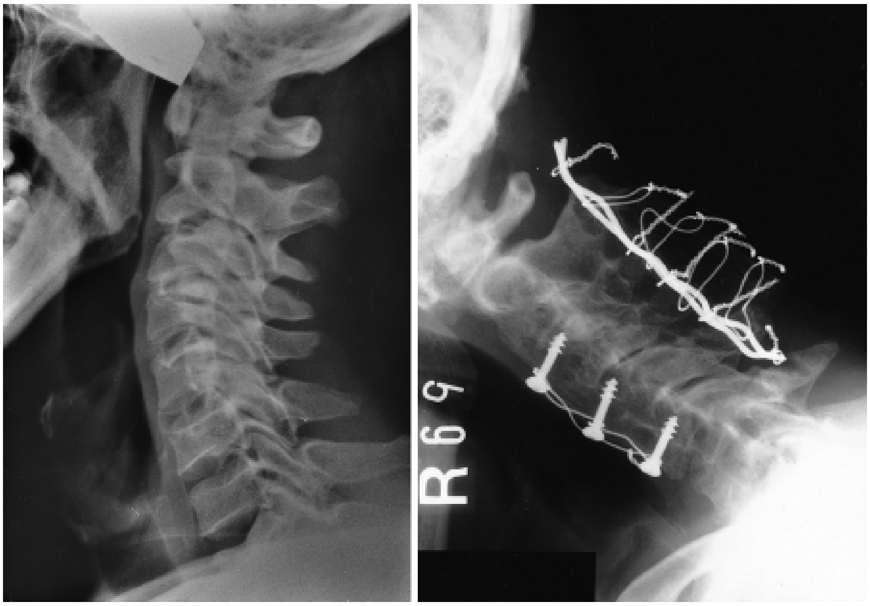


Fig. 5A,B. Roentgenograms in case 5. **A** Lateral radiograph showing cervical spondylotic changes at the third and fourth cervical levels. **B** Lateral radiograph in flexed position made 10 years after surgery, revealing slight atlanto-axial subluxation, complete bony union between C2 and C5, and erosion of the spinous process at C6

Weakness of the deltoid muscle 1 year after op. (MMT)	Weakness of the deltoid muscle at follow-up (MMT)	Non-union	Range of motion of disc space above bloc vertebra (preop.//follow-up)	Range of motion of disc space below bloc vertebra (preop.//follow-up)	Retrolisthesis of vertbera level distance (mm)	Atlanto-axial subluxation/ distance of atlanto-dental interval (mm)
4	4	—	9//0	12//5		
4	4	—	8//0	10//5		
5	5	—	5//4	7//9		
4	4	—	12//0	2//0		5
3	3	—	8//10	20//14		7
5	5	—	3//1	11//11		
5	5	—	13//4	15//7		
5	5	—	8//5	6//5		
4	4	—	9//4	14//20		
4	4	—	4//2	17//17	C4 : 3.0	
5	5	—	12//11	10//0		5
5	5	—	1//0	8//2		7
4	4	—	3//4	5//8		
5	5	—	0//5	1//8		
5	5	—	1//3	6//0		
5	5	—	7//10	10//6		
4	4	—	19//14	0//8	C3 : 3.0	

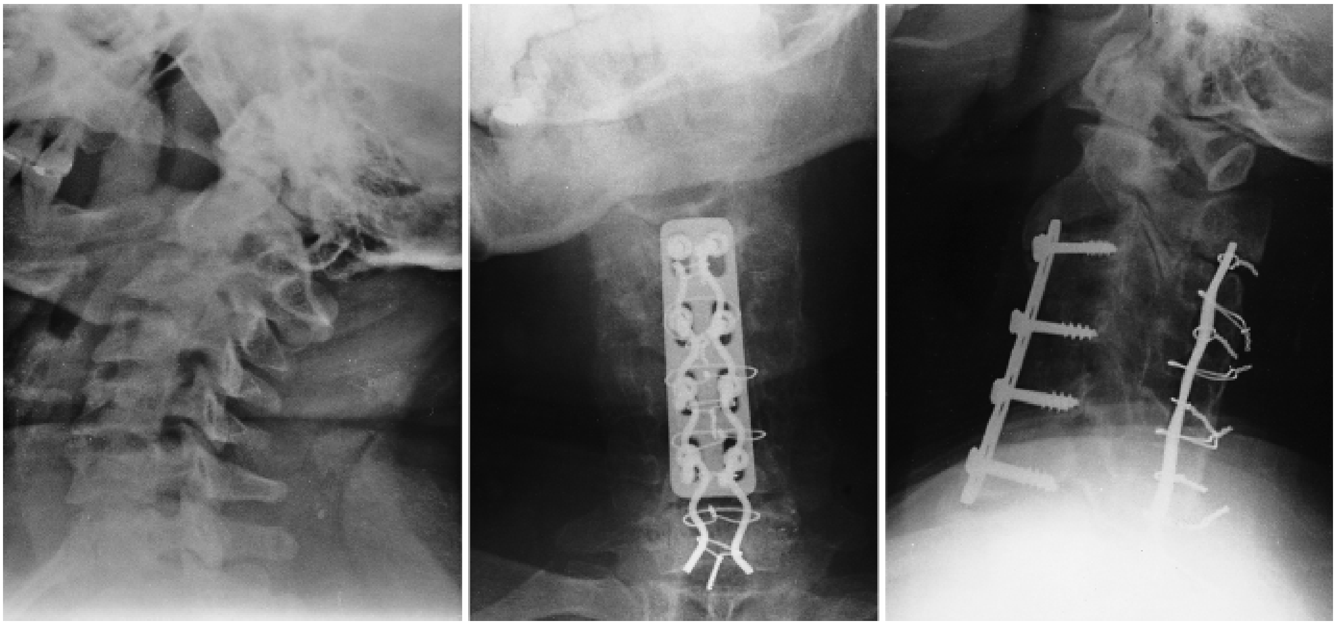


Fig. 6A–C. Roentgenograms in case 2. **A** Lateral radiograph showing spondylotic changes and instability between C3 and C6. **B** Anteroposterior and **C** lateral radiographs made 13 years after surgery, revealing complete bony union between C3 and C6 and remarkable bone formation anteriorly between the second and third cervical levels. The operative result was good

7.0 mm) (Fig. 5). There were no patients with broken or backed out wave-shaped rods, nor were there any fractures of spinous processes. However, in 14 patients, erosion of the spinous processes by the wave-shaped rods was observed on roentgenograms (Fig. 5).

The average diameter of the spinal canal in the sagittal plane was 13.9 mm (range, 11.5–16.0 mm) at the C5 level. In eight patients who had a decline in deltoid muscle strength, the average diameter of the spinal canal in the sagittal plane was 14.9 mm (range, 13.0–16.0 mm), and it was larger than 13.0 mm (range, 11.5–14.0 mm) in the nine patients who did not have a decline in deltoid muscle strength; however there was no significant difference between these diameters.

Discussion

In adults with cerebral palsy, the causes of late dysfunctions, such as dementia, increase in involuntary movements, and articulation disabilities have been reported to be unidentifiable; however, most of them seem to be due to cervical lesions.[16] As has already been pointed out, the most characteristic feature of this pathology is that in most patients the lesions occur at several intervertebral levels and they are often caused by severe instability, mostly between C3 and C4, and between C4 and C5.[6,18] Moreover, dysfunctions such as weakness of the muscles and myatrophy in the upper extremities are also frequently associated with the cervical lesions.[7,10,14]

Myatrophy of the upper extremities is considered to be a disorder of the anterior horn of the spinal gray matter and/or roots of the C5 segment,[2,26] although the reason for its high rate of association with cervical spondylotic myelopathy complicating athetoid cerebral palsy has not yet been identified. In our study, the rate of occurrence of myatrophy was high, and was associated with complete decline in deltoid muscle strength: 8 of our 17 patients demonstrated severe deltoid muscle weakness associated with C5 segmental palsy. The average sagittal diameter of the spinal canal in these patients was 14.9 mm; this was larger than that reported in other studies and in patients with cervical spondylotic myelopathy without cerebral palsy.[19,25]

C5 segmental palsy is considered to be a spinal gray matter or an anterior root and/or nerve root lesion that is caused by instability or compression at the C3/4 and/or C4/5 levels. The resulting spinal canal stenosis frequently causes spinal disorders. However, in the aforementioned eight patients, the symptoms developed without marked spinal canal stenosis, and the anterior root or nerve root lesions had already been present before the patients developed a spinal disorder. Such a condition in the long term was thought to be the cause of the myatrophy or the decline in muscular strength at their first hospitalization.

Since the first report of cervical spondylotic myelopathy in patients with athetoid cerebral palsy, by Anderson et al.[3] in 1962, surgical results have often been poor, because of the unusual operation conditions, such as the patients' involuntary movements and instability, that made conservative treatments ineffective. Levine et al.[12] and McCluer[13] reported on patients who were treated by laminectomy that resulted in deterioration of the disease. Laminectomy improved a spinal disorder only temporarily, but then it frequently caused even more severe instability. This could lead to worsening of symptoms, and therefore we considered this technique inappropriate. Many reports have shown that anterior spinal fusion (with removal of spurs) may lead to good clinical results if bone union could be obtained without such complications as kyphotic deformity, caused by grafted bone loosening or collapse.[7,8,21] But Nakahara et al.,[17] who carried out a long-term study, reported that 4 of 9 operated patients had shown recurrence of myelopathy or radiculopathy.[17] Harada et al.[9] reported similar results: 6 of 19 operated patients experienced the recurrence of symptoms, and these authors pointed out that the spinal stenosis was a critical pathologic factor. The same group also reported on good short-term results of simultaneous surgeries such as laminoplasty and posterior fusion,[9] but other reports indicated that pain and muscular decline in the upper extremities could not be alleviated by laminoplasty.[11] Recently, Murozumi et al.[15] published data on muscular release, and Racette et al.[23] obtained good postoperative results after anterior fusion following the release of muscles with botulinus toxin. These muscle tension-releasing methods may be effective in terms of increasing the bone union rate; however, they should be discussed, especially in regard to their long-term effectiveness, because it is uncertain how long the effects of muscle tension release continue.

We hypothesize that intervertebral instability and cervical malalignment, rather than spinal canal stenosis, affect the development of myelopathy in patients with athetoid cerebral palsy.[1,14,20] Fifteen of the 17 patients in our study did not show spinal canal stenosis of less than 13 mm in the anteroposterior diameter. In fact, our findings support this theory, with data showing that the spinal canal anteroposterior diameter in 17 patients was larger than that usually observed in patients with cervical spondylotic myelopathy. Therefore, considering the aims of stabilizing the cervical spine and correcting malalignment to be the most important for the treatment of this disease, we performed anterior posterior fusion using wave-shaped rods. The anterior posterior fusion technique which was described by Fuji et al.[7] uses interspinous process wiring in accordance with Roger's[22] method. However, with this wiring technique, the appropriate correction of malalignment cannot be achieved as it may be with fusion using wave-shaped rods.[14] Our technique also has the advantage that because both anterior and posterior fixation devices are used, there is no need for rigid external bracing such as a halo vest. Before we developed this technique, we had to exclude from the operative patients those with cervical spondylotic myelopathy whose involuntary movements were very severe.[17] But now we are able to extend our operative treatment indications for those patients also.

Recurrence of the disease was very rare after an average of 8.6 years follow-up, although a few patients demonstrated slight subluxation between C1 and C2.

Conclusion

We conclude that anterior posterior fusion, using wave-shaped rods, is an effective surgical technique for the treatment of cervical spondylotic myelopathy complicating athetoid cerebral palsy.

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