

Factors Affecting Postoperative Complications and Outcomes of Cervical Spondylotic Myelopathy with Cerebral Palsy : A Retrospective Analysis

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Objective : Cervical surgery in patients with cervical spondylotic myelopathy (CSM) and cerebral palsy (CP) is challenging owing to the complexities of the deformity. We assessed factors affecting postoperative complications and outcomes after CSM surgery in patients with CP.

Methods : Thirty-five consecutive patients with CP and CSM who underwent cervical operations between January 2006 and January 2014 were matched to 35 non-cerebral palsy (NCP) control patients. Postoperative complications and radiologic outcomes were compared between the groups. In the CP group, the Japanese Orthopaedic Association score; Oswestry neck disability index; modified Barthel index; and values for the grip and pinch, Box and Block, and Jepsen-Taylor hand function tests were obtained pre- and postoperatively and compared between those with and without postoperative complications.

Results : Sixteen patients (16/35%) in the CP group and seven (7/35%) in the NCP group ($p=0.021$) had postoperative complications. Adjacent segment degeneration ($p=0.021$), postoperative motor weakness ($p=0.037$), and revisions ($p=0.003$) were significantly more frequent in the CP group than in the NCP group; however, instrument-related complications were not significantly higher in the CP group (7/35 vs. 5/35, $p=0.280$). The number of preoperative fixed cervical deformities were significantly higher in CP with postoperative complications (5/16 vs. 1/19, $p=0.037$). In the CP group, clinical outcomes were almost similar between those with and without postoperative complications.

Conclusion : The occurrence of complications during the follow-up period was high in patients with CP. However, postoperative complications did not significantly affect clinical outcomes.

Key Words : Cervical spondylotic myelopathy · Cerebral palsy · Postoperative complications · Kyphosis · Pseudoarthrosis.

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INTRODUCTION

Patients with cerebral palsy (CP) demonstrate involuntary and repetitive neck movements and are more likely to develop cervical spondylotic myelopathy (CSM)^{1,5,6,10,18}. Because CSM progresses in the 30–40-year age range, individuals with CP often develop various neurological defects later in life. The primary pathologic factors that lead to serious disability include compression against neural elements caused by canal stenosis from excessive spondylotic changes and severe dynamic instability of the spine induced by sustained involuntary movements and malalignment of the cervical spine¹⁶. Typically, conservative treatments are ineffective; patients often require surgical treatment². The most important surgical objectives for such patients are adequate decompression of the spinal cord and nerve roots, stabilization of the cervical spine, and alignment correction^{11,12,15}. Recent advances in medical technology have led to the development of improved internal fixation methods, which promote stronger initial mechanical stability resulting from anterior plating or posterior screw fixation⁷. CSM surgery remains challenging, owing to the complexity of the deformity^{8,20,27,28}. Additionally, involuntary neck movements after surgical treatment can worsen neurological symptoms or further complications associated with surgical implants, frequently leading to poor long-term prognoses¹².

We retrospectively investigated the prevalence of various postoperative complications and outcomes resulting from CSM surgery in patients with CP and compared the results with those of non-cerebral palsy (NCP) patients with similar surgical treatments.

MATERIALS AND METHODS

All procedures were performed in accordance with the ethical standards of the Institutional Review Board of Severance Hospital, Yonsei University College of Medicine (approval number : 2016-1447-001). The requirement for informed consent was waived because of the retrospective design of this study.

Patients

Between January 2006 and January 2014, we retrospectively identified 77 patients with CP diagnosed in the Department

of Rehabilitation Medicine and Neurosurgery and CSM who underwent surgeries using cervical anterior (anterior cervical discectomy and fusion or anterior cervical corpectomy and fusion), posterior (posterior decompressive laminectomy and lateral mass screw or pedicle screw fixation), or combined (anterior and posterior) approach in our institute due to intractable pain or progressive weakness. The surgical method and implantation levels were determined by evaluating the extent of the correlation between radiological findings and clinical symptoms. Inclusion criteria were as follows : age >18 years with CP and CSM; subaxial cervical spine lesions; intractable pain or progressive weakness and baseline modified Japanese Orthopaedic Association (mJOA) score \leq 18; follow-up period >2 years; and complete medical record of clinical and radiologic outcomes in the follow-up period. Exclusion criteria were as follows : infections, autoimmune state, tumors, or other pathologic conditions; follow-up period <2 years; and lack of clinical and radiologic outcome data because of poor image quality or follow-up loss. We excluded 42 patients with a follow-up period <2 years or missing clinical and radiologic outcome data. A total of 35 consecutive patients with CP and CSM who underwent cervical surgeries were ultimately included. Regardless of the type of CP before surgery, patients who did not have complications wore the postoperative orthosis for up to 3 months. Patients with complications that were found within 3 months postoperatively wore the orthosis for an additional 2–3 months depending on their condition.

Patient demographics included cervical spine alignment (C0–2 and C2–7 Cobb angle, C2–7 sagittal vertical axis [SVA], chin brow vertical angle [CBVA], and T1 slope [T1S] minus the C2–7 Cobb angle) and postoperative complications. We paired the participants with 35 age-, sex-, and operation level-matched NCP patients who also underwent cervical surgeries owing to CSM.

Data collection

Relevant patient data, including disease history, physical examination findings, and radiological outcomes, were obtained from medical records.

Radiologic outcomes

Plain radiographs, including anterior-posterior and lateral views, were obtained before and 2 years postoperatively, to assess cervical alignment. The C0–2 Cobb angle (neutral posi-

tion), C2–7 Cobb angle (cervical lordosis, CL) (neutral, flexion, and extension positions), T1S, C2–7 SVA, and CBVA were determined. The C0–2 Cobb angle was measured as the angle between the McGregor line and lower endplate of the C2 vertebra (positive values indicate lordosis and negative values indicate kyphosis between the occiput and C2). CL was defined as the angle between two crossed perpendicular lines extending parallel to the inferior endplate of C2 and C7 on a standing lateral radiograph of the cervical spine. The C2–7 SVA was defined as the distance between a plumb line dropped from the center of C2 (or dens) and the posterosuperior aspect of C7. The T1S was measured as the angle between a horizontal plane and a line parallel to the superior T1 endplate. We also evaluated T1S minus CL (T1S–CL), which is a cervical analog to the pelvic incidence minus lumbar lordosis mismatch¹⁴. The C2–7 range of motion (ROM) was calculated by subtracting the CL on flexion from that on extension. The CBVA was defined as the angle between a line from the brow to the chin to the vertical.

Postoperative complications

Postoperative complications, including neurologic deteriorations, adjacent segment disease, instrument-related complications, and revision frequency, were recorded until the last follow-up (at least 2 years after surgery). We also investigated each complication according to the follow-up period.

Clinical outcomes

All clinical outcome assessments, including the mJOA score (range, 0–21; 0=maximal neurological deficits, 21=no neurological deficits)³⁴, Oswestry neck disability index (NDI) questionnaire (10 questions addressing pain intensity²²), personal care, lifting, reading, headaches, concentration, working, driving, sleeping, and recreation, with higher scores indicating worse outcomes), and the modified Barthel index (MBI; range, 0–100; higher scores indicate greater functional independence), were performed preoperatively and 24 months postoperatively in the CP group. Clinical outcomes were also evaluated by a rehabilitation physician using the following tests: grip and pinch (grasping power), Box and Block (performance while carrying a box and block for 1 minute), and Jebsen-Taylor hand function (time to perform certain tasks, including writing, turning cards, picking up small objects, eating, stacking checkers, and carrying light/heavy cans).

Statistical analysis

Data are reported as means±standard deviations. Group differences in demographic and radiologic parameters were evaluated using the chi-square or Fisher's exact test, as appropriate. The paired t-test was used to evaluate differences in clinical parameters between before and 2 years after surgery. Statistical analyses were performed using IBM SPSS Statistics for Windows, version 19 (IBM Corp., Armonk, NY, USA). $p < 0.05$ was considered significant.

RESULTS

Patient demographics

The demographic and clinical characteristics of the patients (35 in each group) are presented in Table 1. In the CP group, there were 21 patients with dyskinetic CP, five with spastic CP, and nine with mixed CP. The average height ($p=0.006$), weight ($p < 0.001$), body mass index ($p=0.001$), and bone mineral density ($p < 0.001$) were significantly lower, and the mean follow-up duration was significantly longer ($p=0.001$) in the CP group than in the NCP group. Osteoporosis ($p=0.007$) and fixed cervical kyphosis ($p=0.003$) were significantly more frequent in the CP group than in the NCP group. There were no significant differences in age ($p=0.610$), sex ratio ($p=1.000$), comorbidities (hypertension, $p=0.690$; diabetes, $p=0.551$), and surgical approach ($p=0.075$) between the groups.

Radiologic outcomes

Preoperative and 2-year postoperative cervical sagittal alignment parameters are summarized according to groups in Table 2. During the preoperative period, the CL on flexion was significantly lower in the CP group than in the NCP group ($p=0.008$). The T1S ($p=0.035$) and C2–7 ROM ($p=0.011$) were significantly higher in the CP group than in the NCP group; however, the C2–7 SVA ($p=0.245$), CBVA ($p=0.893$), and T1S minus the C2–C7 Cobb angle ($p=0.180$) did not differ significantly. In the postoperative period, the C2–7 Cobb angle on flexion was the only parameter that significantly differed between the groups ($p=0.019$). There was no significant difference in radiologic parameters between the groups before and after surgery.

Table 1. Baseline characteristics

	CP (n=35)	NCP (n=35)	p-value
Type of CP			
Dyskinetic, athetoid	21		
Spastic, diplegic	5		
Mixed	9		
Demographic data			
Age (years)	40.0±9.6	41.2±9.6	0.610
Sex, male	21	21	1.000
Height (cm)	160.8±8.6	167.0±9.5	0.006*
Weight (kg)	53.8±12.1	67.5±11.8	<0.001*
BMI (kg/m ²)	20.8±4.5	24.1±2.9	0.001*
BMD	-1.2±1.1	0.4±1.0	<0.001*
Osteoporosis, T-score <-2.5	5	0	0.007*
HTN	3	4	0.690
DM	1	2	0.551
Follow up duration (months)	57.7	32.3	0.001*
Fixed cervical kyphosis	6	0	0.003*
Approach of surgery			0.075
Ant. approach only	20	27	-
Post. with/without ant. approach	15	8	-

Values are presented as mean±standard deviation or number. *Indicates statistical significance ($p<0.05$). CP : cerebral palsy, NCP : non-cerebral palsy, BMI : body mass index, BMD : bone mineral density, HTN : hypertension, DM : diabetes mellitus, Ant. : anterior, Post. : posterior

Postoperative complications

Postoperative complications are summarized in Table 3. There were 16 patients (45.7%) with postoperative complications in the CP group and seven (20.0%) in the NCP group ($p=0.021$). Although the number of overall neurologic deterioration events did not significantly differ between the groups ($p=0.324$), motor weakness was significantly more frequent in the CP group than in the NCP group ($p=0.037$). The groups did not significantly differ in overall instrument-related complications, including pseudoarthrosis, screw loosening, and screw fracture ($p=0.280$); however, adjacent segment degeneration (ASD; $p=0.018$) and revision events ($p=0.003$) were significantly more frequent in the CP group than in the NCP group. Among those with postoperative complications, only

Table 2. Radiologic outcomes between the CP group and the NCP group

	CP (n=35)	NCP (n=35)	p-value
Pre-op radiologic parameters			
C0–C2 angle, neutral (°)	40.6±17.0	37.8±7.8	0.370
C2–C7 angle, neutral (°)	4.3±24.2	3.0±8.5	0.778
C2–C7 angle, flexion (°)	-31.0±17.3	-19.4±18.1	0.008*
C2–C7 angle, extension (°)	22.3±21.6	18.6±12.6	0.380
T1 slope (°)	26.4±13.8	20.9±5.9	0.035*
T1 slope–C27 angle (°)	22.1±17.3	17.9±6.3	0.180
C2–7 SVA (mm)	26.0±22.7	21.4±6.6	0.245
C2–7 ROM (°)	53.3±24.3	38.0±24.4	0.011*
CBVA (°)	-1.8±16.1	-1.5±5.6	0.893
Post-op radiologic parameters			
C0–C2 angle, neutral (°)	40.7±19.8	40.7±7.6	0.992
C2–C7 angle, neutral (°)	2.2±23.3	5.5±10.7	0.459
C2–C7 angle, flexion (°)	-23.3±15.9	-15.2±12.1	0.019*
C2–C7 angle, extension (°)	13.6±23.0	18.4±10.3	0.270
T1 slope (°)	24.7±13.4	23.8±5.7	0.702
T1 slope–C27 angle (°)	22.5±18.3	18.0±9.6	0.204
C2–7 SVA (mm)	25.6±24.0	18.0±8.9	0.081
C2–7 ROM (°)	36.9±27.3	33.5±16.3	0.535
CBVA (°)	-1.3±11.7	-1.9±6.5	0.790
Variation between pre- and postop radiologic parameters			
C0–C2 angle, neutral (°)	0.1±23.3	2.9±5.9	0.490
C2–C7 angle, neutral (°)	-2.0±18.9	2.4±12.1	0.245
C2–C7 angle, flexion (°)	7.7±19.2	4.3±17.2	0.428
C2–C7 angle, extension (°)	-8.7±23.5	-0.2±13.0	0.067
T1 slope (°)	-1.6±11.0	2.9±7.8	0.052
T1 slope–C27 angle (°)	0.4±18.6	0.1±10.4	0.942
C2–7 SVA (mm)	-0.4±21.7	-3.4±11.1	0.471
C2–7 ROM (°)	-16.4±32.6	-4.5±24.1	0.086
CBVA (°)	0.6±18.5	-0.4±8.4	0.774

Values are presented as mean±standard deviation. *Indicates statistical significance ($p<0.05$). CP : cerebral palsy, NCP : non-cerebral palsy, Pre-op : preoperative, SVA : sagittal vertical axis, ROM : range of motion, CBVA : chin brow vertical angle, Post-op : postoperative

the incidence of neurological deterioration significantly differed between groups ($p=0.027$).

In addition, the occurrence of neurologic deterioration was significantly high in the CP group between 2 and 5 years after

Table 3. Distribution of postoperative complications

	CP (n=35)	NCP (n=35)	p-value
Overall complications	16 (45.7)	7 (20.0)	0.021*
Neurologic deteriorations	7 (20.0)	4 (11.5)	0.324
Pain, sensory change without weakness	1 (2.8)	3 (8.5)	0.293
Motor weakness	6 (17.1)	1 (2.8)	0.037*
ASD	7 (20.0)	1 (2.8)	0.018*
Instruments related complications	7 (20.0)	5 (14.3)	0.280
Pseudoarthrosis, non-fusion	2 (5.6)	2 (5.6)	1.000
Screw loosening or fracture	5 (14.3)	3 (8.5)	0.450
Revision	6 (17.1)	0 (0.0)	0.003*
ASD	3 (8.5)	0 (0.0)	-
Motor weakness	6 (17.1)	1 (2.8)	-
Instrument related complications	1 (2.8)	0 (0.0)	-
Period of complications			
≤Post-op 1 year	35	35	
Overall complications	5 (14.3)	5 (14.3)	1.000
ASD	0 (0.0)	0 (0.0)	-
Neurologic deteriorations	1 (2.8)	3 (8.6)	0.293
Instruments related complications	4 (11.4)	3 (9.6)	0.690
Revision	1 (2.8)	0 (0.0)	0.317
Post-op 1 year to post-op 2 years	35	35	
Overall complications	3 (8.6)	1 (2.8)	0.293
ASD	1 (2.8)	0 (0.0)	0.237
Neurologic deteriorations	1 (2.8)	0 (0.0)	0.237
Instruments related complications	2 (5.6)	1 (2.8)	0.551
Revision	1 (2.8)	0 (0.0)	0.237
Post-op 2 years to post-op 5 years	35	16	
Overall complications	8 (22.9)	3 (18.8)	0.738
ASD	3 (8.6)	3 (18.8)	0.311
Neurologic deteriorations	6 (17.1)	0 (0.0)	0.027*
Instruments related complications	2 (5.6)	0 (0.0)	0.214
Revision	3 (8.6)	0 (0.0)	0.126
>Post-op 5 years	16	7	
Overall complications	4 (25.0)	0 (0.0)	0.071
ASD	4 (25.0)	2 (28.6)	0.858
Neurologic deteriorations	2 (12.5)	0 (0.0)	0.216
Instruments related complications	0 (0.0)	0 (0.0)	-
Revision	2 (12.5)	0 (0.0)	0.216

Values are presented as number (%). *Indicates statistical significance ($p < 0.05$). CP : cerebral palsy, NCP : non-cerebral palsy, ASD : adjacent segment disease, Post-op : postoperative

Table 4. Baseline characteristics according to the presence or absence of complications in the cerebral palsy group

	YC (n=16)	NC (n=19)	p-value
Age (years)	37.95±7.06	42.80±11.83	0.140
Sex, male	12	9	0.096
Height (cm)	159.8±7.4	162.1±10.1	0.447
Weight (Kg)	55.5±14.3	51.5±8.3	0.337
BMI (Kg/m ²)	21.7±5.3	19.6±2.7	0.176
BMD	-1.43±1.01	-1.10±1.13	0.386
Osteoporosis	2	3	0.781
HTN	1	2	0.649
DM	0	1	0.264
Type of CP			0.785
Dyskinetic, athetoid	9	12	
Spastic, diplegic	3	2	
Mixed	4	5	
Preoperative history	1	0	0.206
Fixed cervical deformity	5	1	0.037*
Perioperative Botox injection	12	16	0.498

Values are presented as mean±standard deviation or number. *Indicates statistical significance ($p < 0.05$). YC : yes complication, NC : no complication, BMI : body mass index, BMD : bone mineral density, HTN : hypertension, DM : diabetes mellitus, CP : cerebral palsy

surgery ($p=0.027$).

Clinical and radiologic outcomes in the CP group

A summary of demographics and clinical outcomes in the CP group is clustered and presented according to the presence or absence of complications in Tables 4 and 5. In the complication group, the number of fixed cervical deformity was significantly higher than in those without complications ($p=0.037$). The types of CP were not significantly different between the groups with and without complications ($p=0.785$). The pre- ($p=0.049$) and postoperative JOA scores ($p=0.042$) were significantly less in those with postoperative complications than in those without postoperative complications; however, the JOA recovery rate did not significantly differ ($p=0.892$). No other clinical outcomes significantly differed between those with and without postoperative complications. The preoperative and postoperative radiologic parameters also did not differ significantly (Table 6).

Table 5. Clinical outcomes according to the presence or absence of complications in the cerebral palsy group

	YC (n=16)	NC (n=19)	p-value
Clinical outcome			
Pre-op JOA	11.38±4.60	13.95±2.76	0.049*
Post-op JOA score	13.50±3.67	15.74±2.58	0.042*
JOA recovery rate (%)*	25.30±21.07	26.32±22.61	0.892
Pre-op NDI	39.50±20.64	37.95±17.97	0.813
Post-op NDI	26.75±16.96	28.05±16.71	0.821
Pre-op MBI	42.94±23.22	57.53±24.89	0.084
Post-op MBI	62.19±19.49	72.11±20.50	0.154
Grip and pinch test (kg)			
Pre-op grip	18.01±9.41	18.17±7.99	0.956
Post-op grip	15.06±9.23	16.11±8.99	0.736
Pre-op tip	2.62±1.68	2.74±1.78	0.842
Post-op tip	2.57±1.70	2.22±1.47	0.515
Pre-op lateral	3.88±1.85	4.15±2.18	0.696
Post-op lateral	3.79±1.99	3.62±1.88	0.795
Pre-op palmar	3.42±1.66	3.52±2.18	0.888
Post-op palmar	4.01±1.58	3.68±2.56	0.665
Box & Block test			
Pre-op	26.38±16.09	23.57±15.98	0.734
Post-op	23.50±12.89	24.69±16.40	0.689
Jebsen-Taylor hand function test (seconds)			
Writing, pre-op	54.67±48.68	45.81±27.70	0.505
Writing, post-op	43.61±29.31	39.36±25.81	0.652
Card turning, pre-op	19.10±12.30	17.02±11.16	0.604
Card turning, post-op	14.28±8.42	13.13±9.02	0.701
Moving light objects, pre-op	12.35±10.82	12.74±11.83	0.919
Moving light objects, post-op	9.44±3.96	9.24±6.55	0.917
Moving heavy objects, pre-op	9.90±7.37	10.43±7.75	0.839
Moving heavy objects, post-op	9.40±7.06	9.02±6.69	0.873
Stacking checkers, pre-op	22.09±14.37	19.43±13.04	0.575
Stacking checkers, post-op	19.22±14.93	15.85±13.80	0.493
Simulated feeding, pre-op	23.07±14.73	27.14±18.72	0.487
Simulated feeding, post-op	23.29±16.75	21.36±14.72	0.719
Placing small objects, pre-op	36.15±42.43	24.85±18.50	0.301
Placing small objects, post-op	26.20±19.94	18.46±11.15	0.158

Values are presented as mean±standard deviation. *JOA recovery rate (%) : [(preoperative JOA score – postoperative JOA score) / (21 – preoperative JOA score)] × 100. YC : yes complication, NC : no complication, Pre-op : preoperative, JOA : Japanese Orthopaedic Association, Post-op : postoperative, NDI : Neck disability index; MBI : modified Barthel index

Table 6. Radiologic outcomes according to the presence or absence of complications in the cerebral palsy group

	YC (n=16)	NC (n=19)	p-value
Pre-op radiologic parameters			
C0–C2 angle, neutral	43.76±20.33	38.01±13.61	0.326
C2–C7 angle, neutral	2.45±24.48	5.77±24.45	0.691
C2–C7 angle, flexion	-31.68±15.46	-30.44±19.08	0.836
C2–C7 angle, extension	19.41±22.37	24.78±21.22	0.472
T1 slope	28.15±13.56	24.88±14.14	0.493
T1 slope–C27 angle	25.70±18.21	19.11±16.45	0.269
C2–7 SVA (cm)	28.29±26.97	24.18±19.01	0.601
C2–7 ROM	51.09±29.12	55.23±20.02	0.624
CBVA	-3.93±14.00	-0.08±17.81	0.488
Post-op radiologic parameters			
C0–C2 angle, neutral	41.64±20.47	39.93±19.77	0.803
C2–C7 angle, neutral	-0.28±19.59	4.36±26.34	0.565
C2–C7 angle, flexion	-21.38±14.94	-24.85±16.92	0.527
C2–C7 angle, extension	11.21±17.82	15.66±26.96	0.577
T1 slope	24.70±12.74	24.76±14.31	0.989
T1 slope–C27 angle	24.97±18.49	20.40±18.43	0.471
C2–7 SVA (cm)	25.87±29.27	25.44±19.38	0.959
C2–7 ROM	32.59±25.17	40.51±29.06	0.400
CBVA	-0.93±13.55	-1.53±10.23	0.881

Values are presented as mean±standard deviation. YC : yes complication, NC : no complication, Pre-op : preoperative, SVA : sagittal vertical axis, ROM : range of motion, CBVA : chin brow vertical angle, Post-op : postoperative

DISCUSSION

CP is a movement disorder caused by a non-progressive abnormality appearing in the perinatal period. Lack of control and/or spasticity of truncal muscles frequently lead to spinal degenerative diseases or deformities, such as CSM and scoliosis. Early degeneration onset and malalignment of the spine have been reported in patients with CSM and CP. Owing to repetitive and unusual movements associated with this condition, the pathophysiology of CSM accelerates in patients with CP, and inexplicable changes or deterioration of neurological function requiring surgical treatments can occur^{19,21}. CSM surgery for patients with CP remains challenging because of perioperative instrumentation failure caused by the patient's repetitive involuntary neck movements, complicated cervical spine deformities, and comorbidities. Several studies have reported

postoperative complications in patients with CSM without CP or scoliosis with CP^{24,33}. However, there exist only a few reports regarding postoperative complications in patients with CSM and CP. Current and accurate information regarding major complications following cervical surgeries for significant CSM in patients with CP is important for weighing the costs and benefits of corrective surgery for this population.

In previous studies, complication rates in NCP patients who underwent cervical surgery due to CSM were 9–30%^{17,23,26,31}. Yaszay et al.³³ reported a rate of 36% for postoperative major complications (e.g., wound infections, pulmonary issues, instrument-related complications) in patients with CP with >2 years of follow-up after spinal surgery, with a spine-related reoperation rate of 14.0%. Samdani et al.²⁴ reported a 39% complication rate in 127 patients with CP who underwent spinal surgery. In our study, the overall postoperative complication rate for CSM surgery was significantly higher in patients with CP than in NCP patients. Additionally, there were significant group differences in terms of postoperative motor weakness, ASD, and revision events. There may be several reasons for these differences. Excessive involuntary neck motion induced by CP, in association with vertebral slippage anteriorly or posteriorly after cervical fusion, could exaggerate segmental and kyphotic instability. Moreover, these involuntary movements, as well as the abnormal cervical spinal alignment due to an imbalance in cervical muscle tone and compression of neural elements by severe cervical spondylotic changes, can cause rapid and additional degenerative changes and subsequent neurological deficits¹². In our study, six out of 35 patients in the CP group developed motor weakness. In three patients, motor weakness was caused by ASD, and in the other three patients, it was caused by progressive kyphosis. Some studies have reported that cervical ASD occurs in approximately 3% of NCP patients. However, Azuma reported that ASD occurred in 30% of patients with CP with a follow-up period >10 years after cervical fusion for CSM. Likewise, in our study, ASD occurrence was significantly more common in the CP group than in the NCP group. However, instrument-related complications, such as pseudoarthrosis and screw fractures or loosening, did not significantly differ between CP and NCP groups. We hypothesize that improvement in surgical instrument and principles, leading to strong initial mechanical stability as a result of anterior plating or posterior screw fixation, contributed to these favorable results. Additionally, our results

indicate that complications such as instrument failure or non-fusion may not be considered as strong as once thought when considering cervical surgery for CSM in patients with CP.

In our study, risk factors for postoperative complications in CSM with CP included fixed cervical deformity. This result was likely because among patients with CP, surgical invasiveness, need for surgical release, and utilization of osteotomies significantly increased in those with fixed cervical kyphosis than in those with semi-rigid or flexible kyphosis. In patients with CP, fixed cervical kyphosis is often accompanied by severe degenerative changes; thus, surgical treatment of such patients is challenging and requires invasive surgery to correct and stabilize the deformity while decompressing neural elements and restoring sagittal alignment^{11,13,15}. Hence, patients with CSM and CP tend to undergo staged operations or combined approaches. Scheer et al.²⁵ reported significant differences in complication rates for different approaches (anterior approach, 27.3%; posterior approach, 68.4%; combined approach, 79.3%). In the present study, many patients in the CP group required posterior or combined approaches (posterior with an anterior approach or anterior with a posterior approach), but there were no significant differences in surgical approaches between the CP and NCP groups. This is one of the reasons for the high postoperative complication rate.

Apart from the incidence of complications, the clinical outcome is another important factor used to assess the postoperative prognosis. Demura et al.³ showed that the 10-year Barthel index and JOA score after cervical surgery significantly improved (by 36% and 31%, respectively) in 14 patients with CSM and CP. Additionally, Watanabe et al.²⁹ reported that the JOA score improved from 8.3 points preoperatively to 10.9 points by the final follow-up (mean, 58 months) in patients with cervical disorder-associated CP who underwent posterior fusion using cervical pedicle screws; however, one-third of the patients were unable to walk at the final follow-up owing to myelopathy progression. Other studies have shown that postoperative clinical outcomes for patients with cervical myelopathy and CP are relatively poor owing to unfavorable conditions (severe cervical spinal deformity and intervertebral instability associated with involuntary neck movements)^{2,32}. We compared patients with CSM and CP, according to the presence or absence of postoperative complications, to determine the trends in outcomes before and after surgery. We found that JOA scores before and after surgery significantly differed

between those with and without complications; however, the JOA recovery rate, as well as NDI and MBI scores, did not significantly differ. Some studies have reported the imitations of JOA and Barthel index for the evaluation of patients with involuntary movements, as in CP^{4,9)}. Therefore, additional indicators were needed for an accurate evaluation of this patient group³⁰⁾. Accordingly, we assessed the CP group using the functional indicators used in the field of rehabilitation medicine specializing in the evaluation of upper limb function for multilateral assessments (grip and pinch, Box and Block, and Jebsen-Taylor hand function tests). However, no significant differences were found between those with and without postoperative complications. Based on our results, it is not necessary to delay or cancel surgical treatments for patients with CSM and CP due to vague concerns of perioperative complication. Instead, careful consideration of the surgical methods and levels is required, and multidisciplinary consultations should be considered in the treatment of these patients.

Our study has a few key limitations. First, this was a retrospective, uncontrolled drawback study with small sample size. Moreover, the average follow-up period was longer in the CP group than in the NCP group, which was likely to cause selection bias in terms of the complication rate. Second, the rehabilitation medicine clinical scales were only applied to the CP group, and we did not conduct a multifaceted comparison of the clinical courses between the CP and NCP groups. Third, the instrumented level was included while matching the control group (NCP), but there was a fundamental difference in the surgical strategies between the CP and NCP groups. Additionally, the optimal prevention strategy for postoperative complications remains unclear; thus, additional studies are required. Third, the statistical analysis was limited, as this study was designed using a simple matching method for age, sex, and surgical range.

CONCLUSION

The incidence of overall complications, particularly ASD, postoperative motor weakness, and revision events, was higher in the CP group than in the NCP group during the follow-up period (at least 2 years). Risk factors for postoperative complications of CSM with CP included fixed cervical deformity. Instrument-related complications were not significantly different

between groups. In the CP group, there were no differences in clinical outcomes between those with and without postoperative complications. These results suggest that postoperative complications may not be considered as a concern when deciding on surgical treatment for CSM in patients with CP.

CONFLICTS OF INTEREST

No potential conflict of interest relevant to this article was reported.

INFORMED CONSENT

This type of study does not require informed consent.

AUTHOR CONTRIBUTIONS

Conceptualization : HCK, YH, SRC

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Formal analysis : HCK, YH, HJ

Methodology : HCK, HJ, YHJ, SP, SBA

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References

1. Anderson WW, Wise BL, Itabashi HH, Jones M : Cervical spondylosis in patients with athetosis. **Neurology** **12** : 410-412, 1962
2. Azuma S, Seichi A, Ohnishi I, Kawaguchi H, Kitagawa T, Nakamura K : Long-term results of operative treatment for cervical spondylotic myelopathy in patients with athetoid cerebral palsy: an over 10-year follow-up study. **Spine (Phila Pa 1976)** **27** : 943-948; discussion 948, 2002
3. Demura S, Kato S, Shinmura K, Yokogawa N, Yonezawa N, Shimizu T, et al. : More than 10-year follow-up after laminoplasty and pedicle screw fixation for cervical myelopathy associated with athetoid cerebral palsy. **Spine (Phila Pa 1976)** **45** : 727-734, 2020
4. Demura S, Murakami H, Kawahara N, Kato S, Yoshioka K, Tsuchiya H : Laminoplasty and pedicle screw fixation for cervical myelopathy associated with athetoid cerebral palsy: minimum 5-year follow-up. **Spine (Phila Pa 1976)** **38** : 1764-1769, 2013
5. Fuji T, Yonenobu K, Fujiwara K, Yamashita K, Ebara S, Ono K, et al. : Cervical radiculopathy or myelopathy secondary to athetoid cerebral palsy. **J Bone Joint Surg Am** **69** : 815-821, 1987
6. Harada T, Ebara S, Anwar MM, Okawa A, Kajiura I, Hiroshima K : The cervical spine in athetoid cerebral palsy. A radiological study of 180 patients. **J Bone Joint Surg Br** **78** : 613-619, 1996
7. Hartmann S, Thomé C, Abramovic A, Lener S, Schmoelz W, Koller J, et al. : The effect of rod pattern, outrigger, and multiple screw-rod constructs for surgical stabilization of the 3-column destabilized cervical spine - a biomechanical analysis and introduction of a novel technique. **Neurospine** **17** : 610-629, 2020
8. Hasler CC : Operative treatment for spinal deformities in cerebral palsy. **J Child Orthop** **7** : 419-423, 2013
9. Jameson R, Rech C, Garreau de Loubresse C : Cervical myelopathy in athetoid and dystonic cerebral palsy: retrospective study and literature review. **Eur Spine J** **19** : 706-712, 2010
10. Kidron D, Steiner I, Melamed E : Late-onset progressive radiculomyelopathy in patients with cervical athetoid-dystonic cerebral palsy. **Eur Neurol** **27** : 164-166, 1987
11. Kim CW, Hyun SJ, Kim KJ : Surgical impact on global sagittal alignment and health-related quality of life following cervical kyphosis correction surgery: systematic review. **Neurospine** **17** : 497-504, 2020
12. 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
13. Lau D, Ames CP : Three-column osteotomy for the treatment of rigid cervical deformity. **Neurospine** **17** : 525-533, 2020
14. Lee CK, Shin DA, Yi S, Kim KN, Shin HC, Yoon DH, et al. : Correlation between cervical spine sagittal alignment and clinical outcome after cervical laminoplasty for ossification of the posterior longitudinal ligament. **J Neurosurg Spine** **24** : 100-107, 2016
15. Lee JJ, Oh SH, Jeong YH, Park SM, Jeon HS, Kim HC, et al. : Surgical strategies for cervical deformities associated with neuromuscular disorders. **Neurospine** **17** : 513-524, 2020
16. Lee YJ, Chung DS, Kim JT, Bong HJ, Han YM, Park YS : Surgical treatments for cervical spondylotic myelopathy associated with athetoid cerebral palsy. **J Korean Neurosurg Soc** **43** : 294-299, 2008
17. Master DL, Son-Hing JP, Poe-Kochert C, Armstrong DG, Thompson GH : Risk factors for major complications after surgery for neuromuscular scoliosis. **Spine (Phila Pa 1976)** **36** : 564-571, 2011
18. McCluer S : Cervical spondylosis with myelopathy as a complication of cerebral palsy. **Paraplegia** **20** : 308-312, 1982
19. Nishihara N, Tanabe G, Nakahara S, Imai T, Murakawa H : Surgical treatment of cervical spondylotic myelopathy complicating athetoid cerebral palsy. **J Bone Joint Surg Br** **66** : 504-508, 1984
20. Piazzolla A, Solarino G, De Giorgi S, Mori CM, Moretti L, De Giorgi G : Cotrel-Dubousset instrumentation in neuromuscular scoliosis. **Eur Spine J** **20 Suppl 1** : S75-S84, 2011
21. Polk JL, Maragos VA, Nicholas JJ : Cervical spondylotic myeloradiculopathy in dystonia. **Arch Phys Med Rehabil** **73** : 389-392, 1992
22. Pollard CA : Preliminary validity study of the pain disability index. **Percept Mot Skills** **59** : 974, 1984
23. Rumalla K, Yarbrough CK, Pugely AJ, Koester L, Dorward IG : Spinal fusion for pediatric neuromuscular scoliosis: national trends, complications, and in-hospital outcomes. **J Neurosurg Spine** **25** : 500-508, 2016
24. Samdani AF, Belin EJ, Bennett JT, Miyajiri F, Pahys JM, Shah SA, et al. : Major perioperative complications after spine surgery in patients with cerebral palsy: assessment of risk factors. **Eur Spine J** **25** : 795-800, 2016
25. Scheer JK, Tang JA, Smith JS, Acosta FL Jr, Protopsaltis TS, Blondel B, et al. : Cervical spine alignment, sagittal deformity, and clinical implications: a review. **J Neurosurg Spine** **19** : 141-159, 2013
26. Sharma S, Wu C, Andersen T, Wang Y, Hansen ES, Bünger CE : Prevalence of complications in neuromuscular scoliosis surgery: a literature meta-analysis from the past 15 years. **Eur Spine J** **22** : 1230-1249, 2013
27. Shimokawa N, Sato H, Matsumoto H, Takami T : Complex revision surgery for cervical deformity or implant failure. **Neurospine** **17** : 543-553, 2020
28. Vialle R, Thévenin-Lemoine C, Mary P : Neuromuscular scoliosis. **Orthop Traumatol Surg Res** **99** : S124-S139, 2013
29. Watanabe K, Hirano T, Katsumi K, Ohashi M, Shoji H, Yamazaki A, et al. : Surgical outcomes of posterior spinal fusion alone using cervical

- pedicle screw constructs for cervical disorders associated with athetoid cerebral palsy. **Spine (Phila Pa 1976)** **42** : 1835-1843, 2017
30. Watanabe K, Otani K, Nikaido T, Kato K, Kobayashi H, Yabuki S, et al. : Surgical outcomes of cervical myelopathy in patients with athetoid cerebral palsy: a 5-year follow-up. **Asian Spine** **J11** : 928-934, 2017
 31. Wewel JT, Brahimaj BC, Kasliwal MK, Traynelis VC : Perioperative complications with multilevel anterior and posterior cervical decompression and fusion. **J Neurosurg Spine** **20** : 1-6, 2019
 32. Wong AS, Massicotte EM, Fehlings MG : Surgical treatment of cervical myeloradiculopathy associated with movement disorders: indications, technique, and clinical outcome. **J Spinal Disord Tech** **18 Suppl** : S107-S114, 2005
 33. Yaszay B, Bartley CE, Sponseller PD, Abel M, Cahill PJ, Shah SA, et al. : Major complications following surgical correction of spine deformity in 257 patients with cerebral palsy. **Spine Deform** **8** : 1305-1312, 2020
 34. Yonenobu K, Okada K, Fuji T, Fujiwara K, Yamashita K, Ono K : Causes of neurologic deterioration following surgical treatment of cervical myelopathy. **Spine (Phila Pa 1976)** **11** : 818-823, 1986