anterior shift of the spinal cord (Fig. 3f). In this case, we suggest that the cause of C5 palsy was an excessive anterior shift of the spinal cord and tethering of nerve roots. In addition, we speculate that there was asymptomatic damage of the anterior horn cells in the spinal cord, which had existed before surgery and may have also participated in the development of C5 palsy.

Discussion

Sakaura et al. [12] have reviewed the literature regarding postoperative C5 palsy published from 1986 to 2002. They reported that the incidence of C5 palsy after cervical posterior decompression surgery was 4.7% on average, ranging from 0 to 30.0%. On the other hand, its incidence after ASF for cervical lesions was 4.3% on average, ranging from 1.6 to 12.1%, although the number of reports regarding anterior surgery was smaller than that for posterior surgery. To the best of our knowledge, there have been only five large studies on C5 palsy that analyzed more than 100 cases of cervical anterior surgeries [4, 5, 7, 14, 19]. We have summarized their data in Table 6. According to these studies, the incidence of C5 palsy after anterior surgery differed considerably from 3.2 to 9.1%, depending on the institution where they were conducted. The major difference seems to be because a unified definition of C5 palsy has not yet been clearly made. In the present study, we defined C5 palsy as a deterioration of upper extremity motor function by at least one grade in a standard MMT without aggravation of lower extremity function. According to our criteria, the incidence of C5 palsy after the surgeries described was 8.5% in our institute. When the palsy was restricted to a MMT grade ≤ 2, the incidence was 5.0%. When reporting C5 palsy, therefore, it is essential to define it clearly.

Regarding the correlation between the fused levels and the incidence of C5 palsy, Ikenaga et al. [7] described that

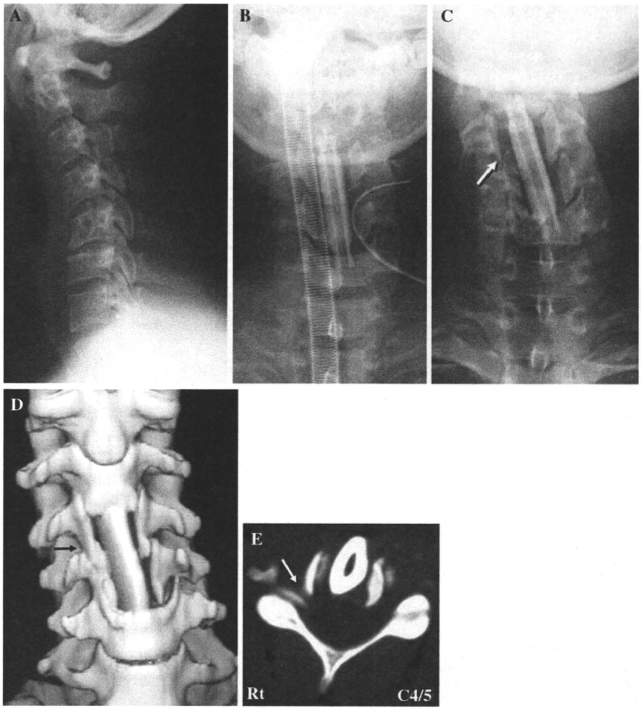

Fig. 2 Case 2. A preoperative lateral cervical radiograph showing kyphotic alignment of the cervical spine (**a**). Anterior–posterior views of cervical radiographs just after anterior corpectomy of C4 and C5 and arthrodesis at C3–6 (**b**) and on the seventh day after surgery (**c**). **c** The lateral tilting angle of the grafted fibula was 14° and the right C4–C5 uncovertebral joint was subluxed (*arrow*). Front view of three-dimensional CT (**d**) and axial CT images at the level of C4–C5 (**e**). CT 8 weeks postoperatively showing a subluxed right C4–C5 uncovertebral joint and stenosis of the right C4–C5 foramen (*arrows*)

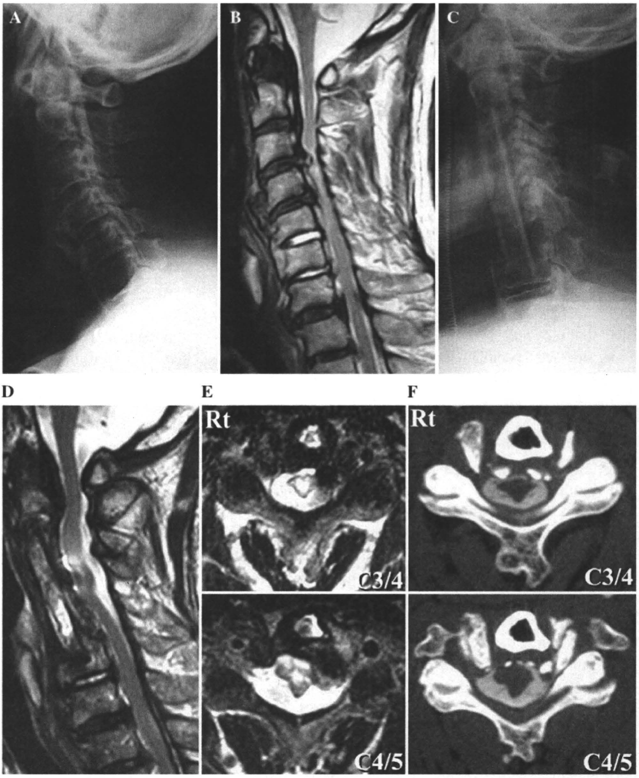

Fig. 3 Case 9. A preoperative lateral cervical radiograph (**a**) showing mixed type OPLL from C1 to C6. A midsagittal T2-weighted MR image (**b**) showing severe compression of the spinal cord and HSCs at C3–C4 and C4–C5 levels. A postoperative lateral cervical radiograph shows anterior corpectomy of C3, C4, and C5 and arthrodesis at C2–C6 (**c**). T2-weighted MR midsagittal (**d**) and axial views at C3–C4 and C4–C5 (**e**) and a CT myelogram (**f**) showing an excessive anterior shift of the spinal cord at C3–C5. **e** HSCs in the gray matter at the C3–C4 and C4–C5 levels

no patient developed C5 palsy after fusion of 1 or 2 levels, and 18 of 362 patients (5.0%) developed the palsy after fusion of 3 or more levels. Greiner-Perth et al. [4] described that 3 of 65 (4.6%) patients developed C5 palsy after fusion of 1 or 2 levels, and 7 of 56 patients (12.5%) developed the palsy after fusion of 3 or more levels. In the present study, we found that 2 of 73 patients (2.7%) developed C5 palsy after fusion of 1 or 2 levels, and 15 of 126 patients (11.9%) developed the palsy after fusion of 3 or more levels. The present results, taken together with those previous reports collectively indicate that the more levels involved in anterior cervical decompression, the more likely the occurrence of C5 palsy.

Previous reports showed that C5 palsy generally had a good prognosis for neurologic and functional recovery. However, irreversible cases of C5 palsy were also reported. Ikenaga et al. [7] reported 7 cases of partial recovery in 18 patients with cases of C5 palsy, and Greiner-Perth et al. [4] reported two cases of partial recovery in ten patients with cases of C5 palsy. In the present study, among 17 patients with cases of C5 palsy, all 7 cases with an MMT grade $\geq$3 at the onset showed complete recovery. However, only five out of ten patients with cases of C5 palsy and an MMT grade $\leq$2 palsy showed complete recovery, three patients recovered incompletely, and two patients remained unchanged at the latest follow-up. For the two palsy patients with no recovery, both had OPLL, and deteriorated to MMT grades 1 and 2 at the onset, and had HSCs on T2-weighted MR images at both C3–C4 and C4–C5 levels. This suggests that, when patients with compression myelopathy and an ossified mass at C3–C4 and C4–C5 levels develop C5 palsy with an MMT grade $\leq$2 after ASF,

Table 6 Incidence and prognosis of C5 palsy after anterior decompression surgery for cervical lesions

Report (year)	No. of patients	No. of cases of palsy (% of patients)	Cases of palsy with MMT grade ≤2 (% of all cases)	1 and 2 levels fusion		3 and more levels fusion		Recovery of the palsy (*n*)	
				No. of cases	Cases of palsy (%)	No. of cases	Cases of palsy (%)	Complete	Incomplete and none
Yonenobu (1991)	204	8 (3.9)	8 (3.9)	ND	0	ND	8	0	8
Saunders (1995)	176	16 (9.1)	ND	20	1 (5.0)	156	15 (9.6)	ND	ND
Greiner-Perth (2005)	121	10 (8.2)	3 (2.5)	65	3 (4.6)	56	7 (12.5)	8	2
Ikenaga (2005)	563	18 (3.2)	7 (1.2)	201	0 (0)	362	18 (5.0)	11	7
Hasegawa (2007)	424	22 (5.2)	ND	ND	ND	ND	ND	ND	ND
Present report	199	17 (8.5)	10 (5.0)	73	2 (2.7)	126	15 (11.9)	12	5

ND Not described

favorable recovery from their palsy may not necessarily be expected. We speculate that certain pre-existing asymptomatic damage to the anterior horn cells in the gray matter of the spinal cord at the C3–C4 and C4–C5 levels may participate in the poor neurological improvement.

Over the past few years, hypotheses regarding the etiology of postoperative C5 palsy have been proposed by many authors. Most of the speculation regarding the etiology is roughly classified into two groups. One hypothesis is that C5 palsy is caused by nerve root damage. Some authors proposed that this might be caused by direct injury to the nerve root [6]. They proposed that surgical instruments could injure neural tissue. However, such a hypothesis cannot explain why many cases of C5 palsy develop several days after the surgery. Others have presumed that tethering of nerve roots might cause C5 palsy as the result of a shift of the spinal cord in association with anchoring of the nerve root [6, 16]. Saunders [14] hypothesized that narrowing of the width of anterior decompression decreased the anterior shift of the spinal cord and the traction of the roots, and consequently prevented the development of radiculopathy after corpectomy. Ikenaga et al. [7] reported the possibility that the extent of anterior dural expansion might have enhanced the incidence of C5 palsy after anterior surgery.

The other hypothesis is that C5 palsy might be caused by a spinal cord disorder. Chiba et al. [1] analyzed C5 palsy after laminoplasty, and proposed that postoperative upper extremity paresis might be caused by a deterioration of the gray matter and proposed that local reperfusion injury in the spinal cord could be the pathomechanism. Hasegawa et al. [5] analyzed C5 palsy after laminoplasty and anterior decompression surgery, and commented that postoperative upper extremity palsy following cervical decompression surgery might result from a transient and localized spinal cord lesion caused by the decompression of a chronic compressive cervical cord disorder. However, both theories regarding the nerve root and spinal cord remain hypothetical because of non-availability of reliable evidence for verifying their proposed hypothetical mechanism.

In the present study, 16 of 17 patients with cases of C5 palsy presented neck and shoulder pain prior to the onset of muscle weakness. This finding supports the theory that postoperative C5 palsy is caused by a certain nerve root lesion. In Case 9, excessive anterior shift of the spinal cord was observed. Because shoulder pain was detected prior to the muscle weakness in this case, it is possible that tethering of C5 nerve roots occurred bilaterally at the posterior edge of excavated vertebra, and that this caused the bilateral C5 palsy. However, the presence of HSCs on T2-weighted MR images at the C3–C4 and C4–C5 levels also provided the possibility of a spinal cord disorder as the etiology of the C5 palsy in this case.

In the present study, it is of particular interest that in all 17 patients with C5 palsy, the most stenotic level of their spinal canal included C3–C4 or C4–C5 levels. In addition, in 12 out of the 17 cases, HSCs were detected at C3–C4 or C4–C5 levels. When restricted to the ten cases of palsy with an MMT grade ≤2, nine cases showed such HSCs. Because all of the 17 patients with C5 palsy had normal muscle power of deltoid and biceps preoperatively, we suggest that asymptomatic damage of anterior horn cells at the gray matter of the spinal cord pre-existed in these cases. Taking all these findings into account, we propose a "double lesion" hypothesis for the development of C5 palsy after ASF for cervical lesion as follows: the pre-existing asymptomatic damage at the anterior horn cells may contribute to the development of postoperative C5 palsy, in combination with nerve root lesions caused by the foraminal stenosis or the excessive anterior shift of the spinal cord after the anterior decompression procedure. Further studies using preoperative and intraoperative electrodiagnostic techniques, such as preoperative EMG and intraoperative neuromonitoring, should provide us with useful information to support this hypothesis.

In conclusion, when patients having spinal cord lesions at C3–C4 and C4–C5 levels undergo multilevel ASF, we should be alert to the possible occurrence of postoperative C5 palsy. To avoid the development of C5 palsy, further improvement of surgical techniques is required to minimize the damage to the nerve roots that may occur subsequent to anterior decompression procedures.

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C5 Palsy after Anterior Cervical Decompression and Spinal Fusion for Cervical Degenerative Diseases

Mitsuhiro Hashimoto[*1], Macondo Mochizuki[*2], Atsuomi Aiba[*2], Akihiko Okawa[*1], Kazuhisa Takahashi[*1], Masashi Yamazaki[*1]

Key words : C5 palsy, cervical spine, anterior surgery

Introduction

Postoperative C5 palsy is one of the major complications after cervical decompression surgery. According to Sakaura et al., the average incidence of C5 palsy was 4.3% (range 1.6-12.1%) in the anterior cervical decompression and spinal fusion (ASF) group, and 4.7% (range 0-30.0%) in the cervical laminoplasty group[8]. However, since fewer studies have analyzed C5 palsy after ASF than after cervical laminoplasty, the clinical and radiological characteristics of C5 palsy after ASF have not been as well defined. The aim of this study was to investigate the incidence and prognosis of C5 palsy after ASF and to discuss the mechanisms behind its development.

Materials and Methods

1. Patient Population

Between 1996 and 2004, consecutive 199 patients underwent ASF for cervical degenerative diseases, including 133 patients with cervical spondylotic myelopathy (CSM), 62 patients with cervical ossification of the posterior longitudinal ligament (OPLL), 16 patients with cervical spondylotic amyotrophy (CSA), 6 patients with cervical spondylotic radiculopathy, and 2 patients with disc herniation. The average age of them was 57.0 years old ranging from 25 to 88 years old. One hundred and forty patients were male and 59 patients were female. The number of fused levels was as follows : one level (33 patients), two levels (40 patients), three levels (46 patients), four levels (71 patients), and five levels (9 patients).

One level fusion was performed by anterior cervical discectomy and fusion, and two or more levels of fusion surgery were performed by anterior cervical corpectomy and arthrodesis. Autologous iliac bone was grafted for one- or two-level fusions and autologous fibula strut was grafted for fusion surgery of three or more levels. In cases where three or more levels were fused, patients were immobilized with a halo vest for 8 weeks after surgery. No additional surgery was performed for the postoperative C5 palsy cases.

2. Clinical Assessments

We defined C5 palsy as deterioration in muscle power of the deltoid or biceps brachii by at least one grade, as assessed by the manual muscle test (MMT), without aggravation of lower extremity function. We retrospectively reviewed their records and evaluated the following clinical measures : onset of radiating neck and shoulder pain, onset of weakness, time course of any MMT grade change, and severity of myelopathy [Japanese Orthopaedic Association (JOA) score].

3. Radiological Assessments

Using lateral cervical radiographs plus computed tomograms (CT) after myelography and magnetic resonance (MR) images, we identified the most stenot-

[*1]Department of Orthopaedic Surgery, Chiba University Graduate School of Medicine [1-8-1 Inohana, Chuo-ku, Chiba 260-8670, Japan]
[*2]Department of Orthopaedic Surgery, Numazu City Hospital

Table 1　Onset and prognosis of C5 palsy

Case no.	Lesion	Levels fused (n)	Impaired muscle	Laterality	MMT grade			Months to recovery	Degree of recovery
					Pre-op.	At onset	At follow-up		
(MMT≤2)									
1	CSM	C4-6(2)	D, B	Rt	5	2	5	3	Complete
2	CSM	C3-6(3)	D, B	Rt	5	2	5	1.8	Complete
3	CSM	C3-6(3)	D, B	Blt	5	2	5	0.8	Complete
4	CSM	C2-6(4)	D, B	Rt	5	2	5	2	Complete
5	OPLL	C3-7(4)	D, B	Lt	5	2	5	1	Complete
6	CSM	C3-7(4)	D, B	Lt	5	2	4	6	Incomplete
7	OPLL	C3-7(4)	D, B	Rt	5	2	4	1.8	Incomplete
8	OPLL	C3-7(4)	D, B	Lt	5	2	4	15	Incomplete
9	OPLL	C2-6(4)	D, B	Blt	5	2	2	NR	No
10	OPLL	C3-7(4)	D, B	Lt	5	1	1	NR	No
(MMT≥3)									
11	CSA	C4-6(2)	D, B	Rt	5	4	5	1	Complete
12	CSA	C3-6(3)	D, B	Lt	5	3	5	1.3	Complete
13	CSM	C3-7(4)	D, B	Rt	5	4	5	2	Complete
14	CSM	C3-7(4)	D, B	Lt	5	4	5	1	Complete
15	CSM	C3-7(4)	D, B	Lt	5	3	5	0.8	Complete
16	CSM	C3-7(4)	D, B	Lt	5	4	5	1	Complete
17	OPLL	C3-7(4)	D, B	Rt	5	3	5	3	Complete

D = deltoid, B = biceps brachii, Rt = right, Lt = left, Blt = bilateral, NR = not recovered

ic level of the spinal column. We also looked for the presence of high signal changes (HSCs) in the spinal cord on preoperative and postoperative T2-weighted MR images.

Results

Overall 17 cases (8.5%) of all 199 cases developed postoperative C5 palsy. The average age of them was 58.5 years old ranging from 26 to 73 years old. Twelve patients were male and 5 patients were female. Stratifying by disease, 9 (7.9%) of the 113 CSM patients, 6 (9.7%) of the 62 OPLL patients, and 2 (12.5%) of the 16 CSA patients developed C5 palsy ; none of the radiculopathy or disc herniation patients developed C5 palsy. Stratifying by number of fused levels, none of the one-level fusion patients, 2 (5.0%) of the 40 two-level fusion patients, 4 (8.7%) of the 46 three-level fusion patients, 11 (15.5%) of the 71 four-level fusion patients and none of the five-level fusion patients developed C5 palsy.

All 17 patients showed some recovery from their myelopathy, the extent of which ranged from 27.6 to 100% (mean 71.2%). Sixteen of these 17 patients presented with radiating neck and shoulder pain prior to their muscle weakness. Pain was recognized 1-7 days (mean 3.6 days) after ASF, and muscle weakness developed 2-23 days (mean 7.2 days) after ASF. No patient had preoperative weakness of the deltoid or biceps brachii. Among the 10 patients with MMT grade ≤2 at the onset, 5 improved to MMT grade 5, 3 improved to MMT grade 4, and 2 with OPLL did not recover at all. Time to maximum recovery ranged from 0.8-15 months (mean 2.8 months) (Table 1). Among all 17 patients, the most stenotic level of the spinal column was either C3-4 or C4-5 disc level ; T2-weighted MR images identified HSCs at C3-4 or C4-5 disc levels in 12 of all 17 patients and 9 of the 10 patients with an MMT grade ≤2 at the onset (Table 2).

Table 2 Radiological chracteristics of 17 patients who developed C5 palsy

Case no.	Most stenotic level	T2W HSC
(MMT≤2)		
1	C4-5	—
2	C4-5, C5-6	C4-5
3	C3-4	C3-4
4	C4-5	C4-5
5	C4-5	C4-5
6	C3-4, C4-5	C3-4, C4-5
7	C4-5, C5-6	C4-5, C5-6
8	C3-4	C3-4
9	C3-4, C4-5	C3-4, C4-5
10	C3-4	C3-4, C4-5
(MMT≥3)		
11	C4-5	—
12	C4-5	—
13	C4-5, C5-6	—
14	C4-5, C5-6	C4-5, C5-6
15	C4-5	C4-5
16	C3-4	C3-4
17	C3-4, C4-5	—

T2W HSC = high signal change in T2-weighted magnetic resonance images

Discussion

There does not seem to be any unified definition of postoperative C5 palsy in previous reports. Hasegawa et al. described that inclusion criteria were deterioration of motor function by at least 1 level in a standard MMT without aggravation of lower extremity function, the appearance of a new sensory disturbance between postoperative day 0 and 2 months after surgery, or both deterioration of motor function and the appearance of a new sensory disturbance between postoperative day 0 and 2 months after surgery, or both deterioration of motor function and the appearance of a new sensory disturbance and that exclusion criteria were pain without change in motor or sensory function, or deterioration of lower extremity function, including tetraparesis[3]. Imagama et al. described that C5 palsy was defined as a paresis of deltoid (MMT grade 0 to 2), with or without involvement of the biceps, but no loss of strength in other muscles[7]. There were reports on postoperative C5 palsy that did not mention the clear definition of C5 palsy[1,2,5,6,9,10]. Only five large studies on C5 palsy have analyzed more than 100 cases of ASF[2,3,6,9,10], among which the incidence of C5 palsy after ASF ranged from 3.2 to 9.1%. One of the major factors responsible for this range seems to be the absence of a unified definition of C5 palsy. In our study, although we observed an 8.5% incidence of postoperative C5 palsy under our criteria, further restricting our definition of palsy to MMT grade ≤2 at the onset yielded an incidence of 5.0%. Providing a clear definition of C5 palsy in one's reports therefore is essential.

Regarding the correlation between the number of fused levels and the incidence of postoperative C5 palsy, Ikenaga et al. reported that no patient developed C5 palsy after fusion of one or two levels, whereas 18 of 362 patients (5.0%) developed C5 palsy after fusion of three or more levels[6]. Similarly, Greiner-Perth et al. reported that 3 of 65 (4.6%) patients developed C5 palsy after fusion of one or two levels in contrast with 7 of 56 patients (12.5%) who developed C5 palsy after fusion of three or more levels[2]. In our study, we identified 2 of 73 patients (2.7%) who developed C5 palsy after fusion of one or two levels *versus* 15 of 126 patients (11.9%) who developed C5 palsy after fusion of three or more levels. Thus our results in conjunction with previous reports collectively indicate that the likelihood of developing C5 palsy increases as ASF involves more corpectomy levels.

Sakaura et al. reported that patients with postoperative C5 palsy generally had a good prognosis for functional recovery[8]. However, irreversible cases of C5 palsy after ASF have been reported. Ikenaga et al. reported 7 cases of partial recovery among 18 patients with C5 palsy[6], and Greiner-Perth et al. reported two cases of partial recovery in ten patients[2]. Among our 17 C5 palsy patients, three patients recovered incompletely, and two patients showed no recovery. For the two palsy patients with no recovery, both had OPLL, and deteriorated to MMT grades 1 and 2 at the onset, had HSCs on T2-weghted MR images at both

C3-4 and C4-5 disc levels. This suggests that, when patients with compression myelopathy and an ossified mass at C3-4 and C4-5 disc levels develop C5 palsy with an MMT grade ≤ 2 after ASF, favorable recovery from their palsy may not necessarily be expected.

Over the past years, various hypotheses have been advanced regarding the etiology of postoperative C5 palsy, which together fall into two basic groups. The first focuses on nerve root lesions as the primary cause, either from direct injury to the nerve root[5] or in association with shifts in the spinal cord and nerve root[6,9]. The other group of hypotheses implicates spinal cord disorders as the source of C5 palsy. For instance, Chiba et al.'s analysis of C5 palsy after cervical laminoplasty indicated that upper extremity paresis might be caused by a deterioration of grey matter, with local reperfusion injury in the spinal cord as the pathomechanism[1].

Sixteen of the 17 patients in our study presented with neck and shoulder pain prior to the onset of muscle weakness, a finding that supports the hypothesis that nerve root lesions may have been the cause of postoperative C5 palsy, just as Saunders stated that a delayed painful weakness of one shoulder was thought to be a C5 radiculopathy[9]. Moreover, the most stenotic level of the spinal canal for all 17 of our C5 palsy patients was C3-4 or C4-5 disc levels. HSCs were detected at the C3-4 or C4-5 disc levels in 12 of these 17 cases and in 9 of the 10 cases with an MMT ≤ 2. These findings indicate that asymptomatic damage of anterior horn cells within the grey matter of the spinal cord was pre-existing in severe postoperative C5 palsy cases. Taking all these findings into account, we would propose a "double lesion" hypothesis for the development of C5 palsy after ASF for cervical lesion as follows : the pre-existing asymptomatic damage at the anterior horn cells may contribute to the development of postoperative C5 palsy, in combination with nerve root lesions after the anterior cervical corpectomy and spinal fusion procedure.

Conclusions

The overall incidence of C5 palsy after ASF among our study patients was 8.5% and higher among those who had multilevel corpectomy and spinal fusion cases. Our findings also suggest that in most of our patients with severe C5 palsy after ASF, pre-existing asymptomatic damage of anterior horn cells at the C3-4 or C4-5 disc levels may have participated in the development of motor weakness in combination with nerve root lesions occurring subsequent to ASF. Our study results thus indicate that clinicians need to be alert to the possible development of postoperative C5 palsy in patients with spinal cord lesions at the C3-4 and C4-5 disc levels who undergo multilevel ASF.

The data listed in Table 1 and Table 2 was cited from the Hashimoto et al.'s published paper[4].

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Etiology of Cervical Spondylotic Myelopathy and Surgical Results of Anterior Cervical Surgery in Elderly Patients

Mitsuhiro Hashimoto*[1], Macondo Mochizuki*[2], Atsuomi Aiba*[2], Akihiko Okawa*[1], Kazuhisa Takahashi*[1], Masashi Yamazaki*[1]

Key words : cervical spondylotic myelopathy, elderly people, anterior cervical surgery

Introduction

Despite the publication of several studies of cervical spondylotic myelopathy (CSM) in elderly patients[1,2,5,6], its etiology and surgical treatment still remain controversial. The purpose of this study was to characterize the etiology of CSM in elderly patients, discuss appropriate surgical treatment based upon its etiology, and evaluate the clinical outcomes of anterior cervical surgery in elderly CSM patients.

Materials and Methods

1. Patient Population

Between 1997 and 2005, 27 CSM patients over 70 years of age (elderly CSM group) and 27 CSM cases under 50 years of age (younger CSM group) were treated at our institute, all of whom underwent anterior cervical surgery. Mean age of the elderly CSM group (16 males, 11 females) was 76 years, ranging from 71 to 85 years. Mean age of the younger CSM group (21 males, 6 females) was 41 years, ranging from 33 to 49 years. Number of fused levels was as follows : among the elderly CSM group, one level (8 patients), two levels (14 patients), three levels (2 patients), and four levels (3 patients) ; among the younger CSM group, one level (7 patients), two levels (6 patients), three levels (4 patients), and four levels (10 patients).

2. Clinical Assessments

OR time, blood loss, and perioperative complications were compared between the elderly and younger CSM groups ; preoperative and postoperative JOA scores were compared with each other within the same group.

3. Radiological Assessments

Diameter of spinal canal in cervical radiographs, olisthesis of each segment in flexion and extension radiographs, and the presence of high signal changes (HSCs) in preoperative and postoperative T2-weighted magnetic resonance (MR) images were compared between the elder and younger CSM groups.

4. Statistical Analysis

The Mann-Whitney *U* test or the Fisher exact probability test was used for statistical analysis. A p-value <0.05 was considered significant, and is identified in the Results section as (*).

Results

With respect to the clinical assessment parameters, among the elderly CSM group, mean JOA score was 8.8 points before surgery *vs.* 12.8 points after surgery (*), and the mean JOA score recovery rate was 49%. Mean OR time was 138 ± 44 minutes, mean blood loss was 59 ± 67m*l*, and no patient required a blood transfusion. Perioperative complications consisted of

*[1]Department of Orthopaedic Surgery, Chiba University Graduate School of Medicine〔1-8-1 Inohana, Chuo-ku, Chiba 260-8670, Japan〕
*[2]Department of Orthopaedic Surgery, Numazu City Hospital

Table 1 Clinical results

	CSM patients over 70 y/o group ($n = 27$)	CSM patients under 50 y/o group ($n = 27$)
JOA score		
Pre op	8.8±2.0	11.8±2.4
Post op	12.8±2.5	15.8±1.2
Post recovery ratio (%)	49±28	75±18
OR time (minutes)	138±44	175±70
Blood loss (m*l*)	59±67	96±121
Complications	Aspiration pneumonia 1 case C5 palsy 1 case	Aspiration pneumonia 1 case C5 palsy 1 case

Table 2 Spinal canal diameter

	CSM patients over 70 y/o group ($n = 27$)	CSM patients under 50 y/o group ($n = 27$)
C3	14.4±2.0mm	13.9±1.3mm
C4	14.2±1.5mm ($p < 0.05$)	13.1±1.4mm
C5	14.3±1.3mm ($p < 0.05$)	13.2±1.5mm
C6	14.6±1.3mm ($p < 0.05$)	13.7±1.4mm
C7	15.0±1.3mm	14.8±1.3mm

Table 3 Olisthesis in each segment

	CSM patients over 70 y/o group ($n = 27$)	CSM patients under 50 y/o group ($n = 27$)
C3-4	2.0±1.5mm ($p < 0.05$)	0.5±0.6mm
C4-5	1.2±1.4mm	1.3±1.0mm
C5-6	0.9±1.3mm ($p < 0.05$)	2.0±0.9mm
C6-7	0mm	0.3±0.5mm

one patient developing an aspiration pneumonia and one patient developing C5 palsy. Among the younger CSM group, mean JOA score was 11.8 points before surgery *vs.* 15.8 points after surgery (*), and the mean JOA score recovery rate was 75%. Mean OR time was 175±70 minutes, mean blood loss was 96±121m*l*, and no patient required a blood transfusion. Perioperative complications consisted of one patient developing an aspiration pneumonia and one patient developing C5 palsy (Table 1).

With respect to the radiological assessment parameters, comparisons of spinal canal diameter between the elderly and younger CSM groups, respectively, were as follows : 14.4±2.0mm *vs.* 13.9±1.3mm at C3, 14.2±1.5mm *vs.* 13.1±1.4 at C4 (*), 14.3±1.3mm *vs.* 13.2±1.5mm at C5 (*), 14.6±1.3mm *vs.* 13.7±1.4mm at C6 (*), and 15.0±1.3mm *vs.* 14.8±1.3 at C7 (Table 2). The incidence of developmental canal stenosis in which spinal canal diameter was less than 13mm in the elderly CSM group was 12 of 27 cases (44%) *vs.* 5 of 27 cases (19%) (*) in the younger CSM group. Comparisons of mean olisthesis between the elderly and the younger CSM groups, respectively, were as follows : 2.0±1.5mm *vs.* 0.5±0.6mm at C3-4 (*), 1.2±1.4mm *vs.* 1.3±1.0mm at C4-5, 0.9±1.3 mm *vs.* 2.0±0.9mm at C5-6 (*), and 0mm *vs.* 0.3±0.5mm at C6-7 (Table 3). Comparisons of the number of patients with HSCs in the spinal cord on T2-weighted MR imaging between the elderly and younger CSM groups, respectively, were as follows : 11 *vs.* 2 patients at C3-4 (*), 14 *vs.* 3 patients at C4-5 (*), 4 *vs.* 16 patients at C5-6 (*), and 0 *vs.* 2 patients at C6-7 (Table 4).

Table 4 Number of patients with high signal changes in T2-weighted MR imaging

	CSM patients over 70 y/o group ($n=27$)	CSM patients under 50 y/o group ($n=27$)
C3-4	11 ($p<0.05$)	2
C4-5	14 ($p<0.05$)	3
C5-6	4 ($p<0.05$)	16
C6-7	0	2

Discussion

Penning placed great importance on retrolisthesis with respect to the pathogenesis of spinal cord compression in spondylotic myelopathy, especially when marked narrowing of the osseous spinal cord in extension is present. Under these conditions, the spinal cord is compressed between bony pincers formed by the anterior aspect of the arch of the inferior vertebra on one side and the posterioinferior aspect of the body of the superior vertebra on the other side, leading him to suggest that this mechanism of compression be referred to as the pincers mechanism[4].

Tani et al. reported that a high incidence (95%) of focal conduction block at C3-4 or C4-5 with normal conduction at C5-6 and C6-7 was characteristic of CSM in elderly people[6]. In another study of elderly CSM patients, Tani et al. found a significant association between degenerative spondylolisthesis and conduction block in the face of a relatively wide spinal canal and concluded that this association indicates the functional importance of degenerative spondylolisthesis in elderly patients with CSM. The authors also consider the higher incidence of both degenerative spondylolisthesis and focal conduction block at the upper cervical levels (C3-4 or C4-5) in this age group to be of clinical interest[5].

Hayashi et al. reported that spondylotic changes such as narrowing of intervertebral discs and osteophytes predominated at the lower disc levels of C5-6 and C6-7, while the upper disc levels of C3-4 and C4-5 exhibited comparatively greater mobility and vertebrolisthesis, especially retrolisthesis in extension. They also reported that pathologic changes at the upper disc levels of C3-4 and C4-5 tended to occur more frequently in aged patients than in younger ones[2]. Hayashi et al. reported in another paper that although their CSM patients did have spinal canal stenosis, the development of myelopathy in their study group did not entirely correlate with AP canal diameter, indicating that other factors besides a narrowed spinal canal might be involved[1].

In our study the cervical spinal canal diameter was wider in the elderly CSM group than that in the younger CSM group, a finding consistent with the results of the study by Hayashi et al. cited above[1]. Moreover our discovery of spondylolisthesis at C3-4 and C4-5 in the elderly group is consistent with the two studies by Tani et al. cited above[5,6]. Consequently we would identify the following radiological findings in the elderly CSM group, compared with the younger CSM group, as characteristic of the elderly CSM group : 1) spinal canal diameter was wider ; 2) spondylolisthesis at C3-4 was more frequently observed ; 3) a higher percentage of patients had HSCs in the spinal cord on T2-weighted MR imaging at C3-4 and C4-5.

Hosono et al. reported that the prevalence of postoperative axial symptoms was significantly higher after laminoplasty than after anterior fusion among their study patients (60% *vs.* 19% ; $p<0.05$). In 18 patients (25%) from the laminoplasty group, the chief complaints after surgery were related to axial symptoms for more than 3 months, whereas in the anterior fusion group, no patient reported having such severe pain after surgery[3]. Indeed, none of the patients in our study experienced severe pain following anterior cervical surgery. These two studies together thus provide evidence that with respect to axial symptoms, anterior cervical surgery appears superior to laminoplasty.

Conclusions

Our study findings identified the following etiologic factors as characteristic of CSM in elderly patients, compared with the younger CSM patients : 1) spinal canal diameter was wider ; 2) spondylolisthesis at C3-4 was more frequently observed ; and 3) a higher percentage of patients had HSCs at C3-4 and C4-5. Anterior cervical surgery for CSM in elderly patients was less invasive in terms of OR time and blood loss and was associated with few perioperative complications and a satisfactory JOA score recovery rate. In light of these findings, anterior cervical surgery is a reasonable procedure for elderly people with CSM. Furthermore, because of its very low rate of postoperative axial symptoms, anterior surgery that does not cause muscle injury may be preferable to laminoplasty.

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Occipitocervical Fusion Has Potential to Improve Sleep Apnea in Patients With Rheumatoid Arthritis and Upper Cervical Lesions

Hiromi Ataka, MD, PhD,* Takaaki Tanno, MD, PhD,* Tomohiro Miyashita, MD, PhD,*
Shiroh Isono, MD, PhD,† and Masashi Yamazaki, MD, PhD‡

Study Design. Case series.

Objective. To analyze factors that contribute to the development of sleep apnea in patients with rheumatoid arthritis (RA) and upper cervical lesions.

Summary of Background Data. No large prospective study has analyzed the association between sleep apnea and upper cervical involvement resulting from RA. Furthermore, only 1 report in the literature describes a case of sleep apnea accompanying rheumatoid vertical subluxation of the odontoid process.

Methods. The authors analyzed 8 consecutive RA patients with upper cervical lesions who underwent occipitocervical (O-C) fusion. The patients were examined with all-night polysomnography before and after surgery. Patients with apnea-hypopnea index values ≥5 were diagnosed to have sleep apnea. O-C2 angles were calculated from cervical radiographs.

Results. All 8 patients were diagnosed as having sleep apnea, and most of their apneic episodes were obstructive in origin. Among the 4 patients with medullary compression, central apneic episodes comprised ≤5% of their respiratory events. Two patients with severe sleep apnea had negative O-C2 angles. Six patients who showed postoperative improvements in their sleep apnea all had positive changes in their O-C2 angles exceeding 5° after surgery. The differences between preoperative and postoperative O-C2 angles were significantly greater in the patients with improvement of sleep apnea than in the patients with worsening sleep apnea.

Conclusion. All our study patients with RA and upper cervical lesions had obstructive-dominant sleep apnea. Negative O-C2 angles may result in upper airway narrowing, increasing the severity of sleep apnea. O-C fusion with correction of kyphosis at the craniovertebral junction has the potential to improve sleep apnea in RA patients.

Key words: sleep apnea, rheumatoid arthritis, craniovertebral junction, O-C2 angle, occipitocervical fusion. **Spine 2010;35:E971–E975**

From the *Department of Orthopaedic Surgery, Matsudo City Hospital, Chiba, Japan; and †Department of Anesthesiology, and ‡Spine Section, Department of Orthopaedic Surgery, Chiba University Graduate School of Medicine, Chiba, Japan.

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Address correspondence and reprint requests to Masashi Yamazaki, MD, PhD, Spine Section, Department of Orthopaedic Surgery, Chiba University Graduate School of Medicine, 1-8-1 Inohana, Chuo-Ku, Chiba 260-8677, Japan; E-mail: masashiy@faculty.chiba-u.jp

Sleep apnea associated with rheumatoid arthritis (RA) was first described by Davies and Conrad in 1983.[1] Since then, several reports have described patients with RA who presented with obstructive sleep apnea, central sleep apnea, or a combination of both.[2–5] Obstructive sleep apnea is associated with a variety of conditions that contribute to upper airway narrowing, such as development of retrognathia secondary to temporomandibular joint destruction.[2–4] Central sleep apnea is caused by dysregulation of the respiratory center located in the brain stem. In patients with RA, this dysregulation can occur if vertical luxation of the odontoid process leads to compression of the brain stem and consequent impairment of the breathing center.[5] Nevertheless, most of those previous reports have focused on upper airway narrowing secondary to temporomandibular joint destruction. No large prospective study in the literature has systematically studied the association between sleep apnea and upper cervical spine involvement resulting from RA. Drossaers-Bakker et al[5] described 1 patient who presented with sleep apnea that accompanied vertical luxation of the odontoid secondary to RA and improved after occipitothoracic fusion.

In the study herein involving 8 patients with RA who underwent occipitocervical (O-C) posterior fusion for upper cervical lesions in our institute, we analyzed morphologic factors that contribute to the development of sleep apnea. We also evaluated postoperative changes in the severity of sleep apnea and their relationship to radiologic changes to assess the potential of O-C fusion to improve sleep apnea.

Materials and Methods

Patient Population

Eight consecutive patients underwent posterior O-C fusion for upper cervical spine involvement secondary to RA at our institute between September 2006 and September 2007 (Table 1). All were women, and the mean age at surgery was 68.4 years (range, 63–75 years). Body mass index ranged from 18 to 22 kg/cm^2; none of the patients were obese. The mean duration of RA was 26.9 years. Six of the 8 patients had the mutilans type of RA. Using the functional classification of the American College of Rheumatology to assess RA severity, we categorized 4 patients as class III and the other 4 patients as class IV. Rheumatoid involvement of the upper cervical spine was radiographically defined as one of, or a combination of the following patterns of instability: anterior atlantoaxial subluxation (AAS), vertical subluxation (VS), or atlanto-occipital joint subluxation. Among our set of patients, 1 had AAS alone, 1 had VS

Table 1. Clinical Characteristics of Eight Patients Who Underwent Occipitocervical Fusion for Upper Cervical Lesions Secondary to Rheumatoid Arthritis

Case No.	Age (yrs)/ Sex	Radiographic Findings	Medullary Compression	Pain Grade		Neurologic Status		AHI		Preoperative Apneic Episodes	
				Preoperative	Postoperative	Preoperative	Postoperative	Preoperative	Postoperative	% Obstructive	% Central
1	72/F	AAS + VS + SS	+	2	0	IIIB	IIIB	40.3	16.4	99.5	0.5
2	72/F	AAS + VS	+	2	0	IIIB	IIIB	45.5	7	99.6	0.4
3	75/F	AAS + VS + SS	−	3	1	II	I	9.5	3.5	100	0
4	69/F	AAS + AOS	−	3	1	I	I	23.5	27.3	82.6	17.4
5	64/F	AAS + VS	−	2	0	IIIA	II	19.4	13.1	97.9	2.1
6	69/F	AAS + VS	−	3	0	IIIA	II	28.1	31.8	92.9	7.1
7	65/F	VS	+	3	0	IIIA	IIIA	15.1	3.5	100	0
8	63/F	AAS	+	3	0	IIIA	II	6	2.3	95	5

AHI indicates apnea-hypopnea index; AAS, atlantoaxial subluxation; VS, vertical subluxation; AOS, atlanto-occipital subluxation; SS, subaxial subluxation.

alone, 1 had AAS and atlanto-occipital joint subluxation, 3 had AAS and VS, and the remaining 2 had AAS and VS along with subaxial subluxation (SS). Overall, 6 patients had VS, either alone or in combination with AAS.

Indications for O-C fusion included intractable pain or neurologic deficit or both with signs of myelopathy. Subjective pain level and neurologic status were determined according to the method of Ranawat *et al.*[6] All patients had moderate or severe neck and occipital pain (grade 2 or 3) before surgery. Seven patients had neural deficits: class II, 1; class IIIA, 4; or class IIIB, 2. All 8 patients were judged to need O-C fusion on the basis of superior migration of the dens, irreducible subluxation, and significant atlanto-occipital pathology. Seven patients had a posterior fusion from the occiput to C2. The eighth patient (case 1), 1 of the 2 subjects with AAS and VS plus SS, had a subaxial extension of the fusion to T1 to treat superior migration of her odontoid process and the SS. All patients experienced relief of their neck and occipital pain after surgery. No patient required opioids before or after surgery for pain control. Neurologic status improved in 4 patients and remained unchanged in the remaining 4 patients; no patient experienced a neurologic decline as a result of surgery (Table 1).

Sleep Studies

All patients underwent all-night polysomnography studies before and 1 to 3 months after surgery with a diagnostic sleep recorder (Stardust II; Philips Respironics, GK., Tokyo, Japan), which included recording of nasal airflow, respiratory effort, pulse oximetry (pulse rate and percentage oxygen in the blood), and body position (supine or nonsupine). Episodes of apnea (total airflow blockage) and hypopnea (partial airflow blockage) were detected by measuring pressure, base airflow, pulse rate, oxygen saturation and desaturation, chest and abdominal effort, and body position changes *via* wires and a belt with built-in sensors.

Respiratory parameters analyzed were as follows: number of obstructive apneic episodes (absence of air flow associated with persistent thoracoabdominal movement lasting ≥10 seconds; Figure 1A); number of central apneic episodes (absence of air flow and thoracoabdominal movement lasting ≥10 seconds; Figure 1B); and number of hypopneic episodes (reduction of air flow by ≥50% with thoracoabdominal movement for ≥10 seconds, or a 3% drop in the oxygen saturation level).[7] The apnea-hypopnea index (AHI) was calculated as the number of abnormal respiratory events per hour. Patients with AHI values ≥5 were diagnosed as having sleep apnea.

Radiographic Assessment

Plain lateral radiographs of the cervical spine in flexion were evaluated to calculate the O-C2 angle and the Redlund-Johnell distance before and after the surgery. The O-C2 angle was defined as the angle between McGregor's line and the inferior endplate of C2 (Figures 2 and 3). A negative O-C2 angle indicates kyphosis at the craniovertebral junction. The Redlund-Johnell distance was defined as the vertical distance between the midpoint of the C2 endplate and McGregor's line (Figure 2). A Redlund-Johnell distance measuring <34 mm in men or 29 mm in women indicated basilar invagination because of VS.[8] The presence or absence of compression of the medulla oblongata was determined from magnetic resonance (MR) images: if T2-weighted sagittal MR images showed disappearance of the ventral subarachnoid space at the level of the foramen magnum, medullary compression was considered to be present. On T1-weighted sagittal MR images, a line was drawn along the ventral side of the medulla oblongata and a second line

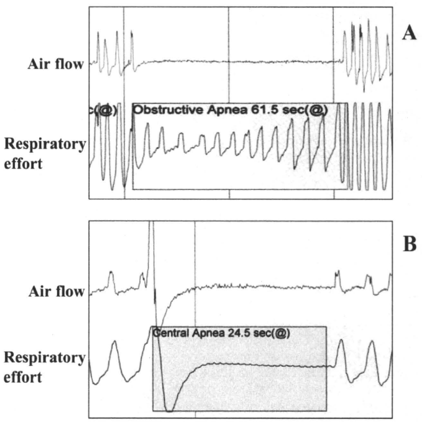

Figure 1. Typical polysomnography recordings showing obstructive apnea **(A)** and central apnea **(B)**. **(A)** Obstructive apnea showing respiratory effort without airflow. **(B)** Central apnea showing the absence of both airflow and respiratory effort.

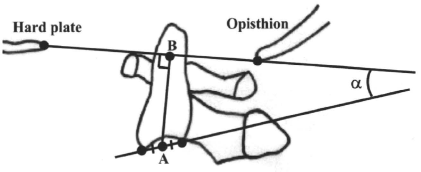

Figure 2. Schematic diagram of radiographic measurements. O-C2 angle: the angle between McGregor's line and the inferior endplate of C2. Redlund-Johnell distance: the vertical distance between the midpoint of the inferior endplate of C2 and McGregor's line.

drawn along the anterior aspects of the upper cervical spinal cord; the cervicomedullary angle was defined as the angle between these 2 lines.[9] Statistical comparisons used the Wilcoxon signed rank test and the Mann-Whitney *U* test. Statistical significance was set at $P < 0.05$.

■ Results

All the patients with RA and upper cervical lesions who underwent O-C fusion were diagnosed as having sleep apnea, with AHI values ranging from 6 to 45.5 (Table 1). Two patients (cases 1 and 2) had severe sleep apnea with AHI ≥30; 4 patients (cases 4, 5, 6, and 7) had moderate sleep apnea (15 ≤ AHI <30); and 2 patients (cases 3 and 8) had mild sleep apnea with AHI <15. For all the 8 patients, most apneic episodes were obstructive in origin, with obstructive apneic episodes comprising >80% of all their respiratory events. Among the 4 patients with medullary compression, central apneic episodes constituted ≤5% of all their respiratory events. Two patients with severe sleep apnea (cases 1 and 2) had negative preoperative O-C2 angles (−26° and −8°), demonstrating severe kyphosis at the craniovertebral junction; overall, preoperative O-C2 angles among the 8 patients ranged from −26° to 12° (Table 2).

Six (cases 1, 2, 3, 5, 7, and 8) of the 8 patients evidenced improvement of their sleep apnea after surgery, 3 of whom (cases 3, 7, and 8) experienced complete recovery (postoperative AHI <5; Table 1). All of these 6 patients who enjoyed postoperative improvements in their sleep apnea had positive changes in their O-C2 angles exceeding 5° after surgery. Figure 3 presents an illustrative example (case 7) of such a positive change in the O-C2 angle after surgery (16°), as demonstrated by a comparison of preoperative (A) and postoperative (B) radiographs. Conversely, the 2 patients (cases 4 and 6) whose sleep apnea did not improve after surgery had minimal or no change in their O-C2 angles after surgery (4° and 0°, respectively). Statistical analysis demonstrated that the differences between preoperative and postoperative O-C2 angles (ΔO-C2 angle = postoperative O-C2 angle − preoperative O-C2 angle) were significantly greater among the patients with postoperative improvement in AHI (cases 1, 2, 3, 5, 7, and 8) than among the patients with postoperative deterioration in AHI (cases 4 and 6). However, ΔR-J distance (postoperative Redlund-Johnell distance − preoperative Redlund-Johnell distance) did not show a significant difference between the patients with postoperative improvement in AHI and the patients with postoperative deterioration in AHI. In 6 of the 8 patients, the cervicomedullary angle was measured before and after surgery. Among these 6 patients, the cervicomedullary angle increased after surgery by 7° to 23° (Table 2).

■ Discussion

All of our study patients with RA and upper cervical lesions who underwent O-C fusion at our institute had sleep apnea before surgery. An epidemiologic study in the United States estimated the prevalence of sleep apnea, defined as an AHI of 5 or higher, as 9% for women

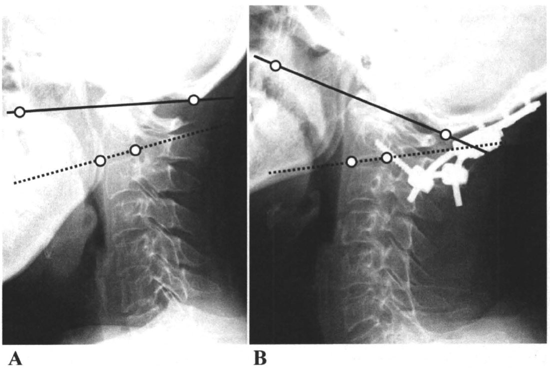

Figure 3. Radiographs from case 7 showing preoperative and postoperative morphology of the craniovertebral junction. **A**, Preoperative lateral radiograph showing kyphosis at the craniovertebral junction: O-C2 angle = 7°. **B**, Postoperative lateral radiograph showing correction of the kyphosis: O-C2 angle = 23°.

Table 2. Pre- and Postoperative Radiologic Measurements

Case No.	O-C2 Angle (°) Preoperative	O-C2 Angle (°) Postoperative	ΔO-C2 Angle (°)	R-J Distance (mm) Preoperative	R-J Distance (mm) Postoperative	ΔR-J Distance (mm)	CMA (°) Preoperative	CMA (°) Postoperative	ΔCMA (°)
1	−26	−12	14	3	8	5	101	123	22
2	−8	1	9	15	24	9	133	147	14
3	8	14	6	26	27	1	166	NA	NA
4	12	16	4	29	31	2	133	147	14
5	10	27	17	26	26	0	145	152	7
6	9	9	0	19	20	1	143	NA	NA
7	7	23	16	23	24	1	136	151	15
8	−14	12	26	37	39	2	143	166	23

ΔO-C2 angle indicates postoperative O-C2 angle − preoperative O-C2 angle; ΔR-J distance, postoperative R-J distance − preoperative R-J distance; CMA, cervicomedullary angle; NA, not assessed; ΔCMA, postoperative CMA − preoperative CMA.

and 24% for men in an apparently healthy middle-aged population.[10] A study of an elderly population aged 65 or more estimated the prevalence of sleep apnea as 19.5% for women and 28% for men.[11] Comparing these estimates with the observation that all our study patients had sleep apnea suggests that the incidence of sleep apnea may be much higher in patients with RA and upper cervical lesions than in healthy populations.

Several previous studies have suggested that destruction of the upper cervical spine by RA, leading to potential compression of the medulla, may be a causative factor for obstructive sleep apnea as well as for central sleep apnea.[3,5] Drossaers-Bakker *et al*[5] described a patient with RA and vertical luxation of the odontoid who developed combined obstructive and central apnea. They hypothesized that the central component of sleep apnea, caused by compression of the brain stem, might reduce muscle tone in the upper airways, thereby contributing to the obstructive component. However, among our 8 study patients, only 2 had pure obstructive sleep apnea; the other 6 had combined obstructive and central apnea even though >80% of their apneic episodes were obstructive in origin. Moreover, for the 4 patients with medullary compression, central apneic episodes still constituted ≤5% of their respiratory events. These findings suggest that compression of the brain stem is not necessarily a major causative factor of sleep apnea in RA patients.

Obstructive sleep apnea is caused by collapse of the pharyngeal airway. The size of the pharyngeal airway is regulated by a precise interaction between the action of upper airway dilator muscles. During sleep, muscle tone throughout the body decreases along with relaxation of the upper airway dilating muscles and narrowing of the upper airways, even in normal subjects.[12] In patients with RA, a reduction in the size of the upper airway can easily occur in association with retrognathia because of temporomandibular joint destruction,[2–4] leading to the development of obstructive sleep apnea.

In an earlier study, we documented that the size of the pharyngeal airway varies with positioning changes of head and neck.[13] Specifically, we observed that neck flexion decreased maximum pharyngeal airway size and increased pharyngeal closing pressure, leading to obstructive sleep apnea. In our study herein, the 2 patients with severe sleep apnea had negative initial O-C2 angles, a finding which suggests that negative O-C2 angles aggravate obstructive sleep apnea. In particular, the O-C2 angle is an indicator of the alignment of the craniovertebral junction. A negative value of the O-C2 angle indicates that the craniovertebral junction has a flexed alignment, resulting in narrowing and increased collapsibility of the upper airway. Miyata *et al*[14] reported that negative changes in the O-C2 angle after O-C posterior fusion in RA patients were associated with the development of dyspnea or dysphagia or both. Their results support the implication of our study findings that a negative O-C2 angle and corresponding flexion of the upper cervical spine may result in upper airway narrowing that increases the severity of obstructive sleep apnea. Their results were also in line with the outcome of our study that the 6 patients who showed improvement or complete recovery from their sleep apnea after craniovertebral fusion were also the patients who had positive changes in their O-C2 angles exceeding 5° after surgery.

Abumi *et al*[15] reported that O-C fusion with correction of kyphosis at the craniovertebral junction causes indirect decompression of the upper spinal cord, which, in turn, could relieve several of the symptoms resulting from upper cervical lesions in RA patients. In our study herein, we measured the cervicomedullary angle in 6 patients and found that this angle increased after surgery in all 6 patients, indicating indirect decompression of their spinal cords occurred after O-C fusion. However, the small number of patients in our study precluded us from determining whether indirect decompression of their spinal cords was associated with a significant improvement in their sleep apnea. A multi-institutional study with a large number of patients, thus, will be needed to statistically evaluate this possible association between indirect decompression of the spinal cord and subsequent improvement in sleep apnea.

Previous reports have shown that obstructive sleep apnea and central sleep apnea commonly occur in patients undergoing long-term opioid therapy and that these sleep disorders improve after opioid withdrawal.[16,17] These findings have led to the hypothesis that in patients experiencing a relief of neck and occipital pain after O-C fusion, which enables them to reduce or fully stop their use of opioids, postoperative improvement in their sleep apnea may be attributable at least in part to the opioid withdrawal. This hypothesis is not applicable to our study subjects because none of our patients received opioids before or after surgery. However, further studies will be required to examine the influence of other analgesics, including steroids, on the development of sleep apnea in RA patients.

One limitation of our study was that we did not evaluate temporomandibular joint destruction. Because previous studies have reported that patients with RA and upper cervical involvement often had destruction of the temporomandibular joints,[4,18] it is plausible that some or even most of the 8 patients in our study also may have suffered destruction of their temporomandibular joint, which could have resulted in upper airway narrowing. However, because 6 of our 8 patients showed improvement in their obstructive-dominant sleep apnea after O-C fusion, even without receiving treatment for temporomandibular joint destruction, our study findings suggest that the development of obstructive sleep apnea is more closely related to O-C2 alignment than to temporomandibular joint destruction.

All our study patients with RA and upper cervical lesions who underwent posterior O-C fusion had a high prevalence of obstructive-dominant sleep apnea. Although various factors may cause upper airway narrowing in patients with obstructive sleep apnea, our study results suggest that O-C2 angle has a major impact on its development and aggravation. Although our sample size was relatively small, our study results suggest that O-C fusion with correction of kyphosis at the craniovertebral junction has the potential to improve sleep apnea in patients with upper cervical involvement secondary to RA.

■ Key Points

- All 8 study patients with rheumatoid arthritis and upper cervical lesions who underwent occipitocervical (O-C) fusion were diagnosed as having obstructive-dominant sleep apnea.
- Two patients with severe sleep apnea had negative O-C2 angles.
- Six patients who showed postoperative improvements in their sleep apnea all had positive changes in their O-C2 angles exceeding 5°.
- O-C fusion with correction of kyphosis at the craniovertebral junction has the potential to improve sleep apnea.

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Original article

Surgical correction of spinal deformity in patients with congenital muscular dystrophy

Masashi Takaso[1], Toshiyuki Nakazawa[1], Takayuki Imura[1], Takamitsu Okada[1], Masaki Ueno[1], Wataru Saito[1], Kazuhisa Takahashi[2], Masashi Yamazaki[2], and Seiji Ohtori[2]

[1]Department of Orthopaedic Surgery, Kitasato University, School of Medicine, Kitasato 1-15-1, Minami Sagamihara, Kanagawa 252-0375, Japan
[2]Department of Orthopaedic Surgery, Chiba University, Graduate School of Medicine, Chiba, Japan

Abstract

Background. Congenital muscular dystrophy (CMD), among the myopathic disorders is one form of flaccid neuromuscular disorder (NMD). Patients with NMD frequently develop progressive spinal deformity. For NMD patients who have a severe spinal deformity, sitting is often difficult and is accompanied by pain and breakdown of the skin. Spinal deformity surgery in these patients has been highly effective in stabilizing the spine, maintaining upright, comfortable sitting balance, and improving patients' quality of life. However, many studies have reported significant rates of peri/postoperative complications in these patients. To our knowledge, there has been no study on the results of spinal deformity surgery in patients with CMD. The purpose of this study was to review the clinical and radiological results of spinal deformity surgery in this group of patients with CMD.

Methods. Between 2004 and 2007, a total of 10 CMD patients underwent scoliosis surgery. There were three patients with Fukuyama CMD, three with Ullrich CMD, and 4 with nonsyndromic CMD (merosin-negative). They were nonambulatory. All the patients had standard posterior spinal fusion and pedicle-screw-alone fixation from T3 or T4 to L5 for spinal deformity. Our inclusion criteria required that each patient (1) had considerable difficulty with sitting balance and pain or breakdown of the skin due to scoliosis; (2) was able to ventilate his or her lung autonomously; (3) was not ventilator-dependent; and (4) did not have cardiac failure. Sufficient informed consent was important, and the decision to perform surgery was made by the patient/family with sufficient preoperative informed consent. Patients were trained with inspiratory muscle training (IMT) using an inspiratory muscle trainer (Threshold IMT) for 6 weeks prior to surgery Cardiac function was assessed preoperatively. Pulmonary function tests were performed preoperatively and postoperatively. Radiographic assessments were performed on sitting anteroposterior (AP) and lateral radiographs. These assessments were made periodically. The Cobb angles of the curves and spinal pelvic obliquity (SPO) on the coronal plane, thoracic kyphosis, and lumbar lordosis were measured. The preoperative AP radiograph and side-bending films were examined to determine flexibility. Patients' and parents' satisfaction were surveyed by a self-completed questionnaire at the last follow-up.

Results. Percent forced vital capacity (%FVC) increased from a mean of 30% before IMT to a mean of 34% the day before surgery. The preoperative scoliosis was 75° (range 61°–95°). The scoliotic curvature on preoperative side-bending films was 19° (range 11°–28°). All patients were extubated on the day of surgery. No patients developed cardiac or respiratory complications. The scoliotic curvature was 18° (range 10°–25°) immediately after surgery, and 19° (range 12°–27°) at the last follow-up. The pelvic obliquity improved from a mean of 17° (range 14°–20°) preoperatively to a mean of 6° (range 4°–9°) postoperatively and to 7° (range 4°–10°) at the last follow-up. Balanced sitting posture was achieved and maintained. On the sagittal plane, good reconstruction of sagittal plane alignment was recreated and maintained. There were no major complications or deaths. All patients/parents completed the outcome satisfaction questionnaire. Eight patients/parents were very satisfied and two were satisfied.

Conclusions. Pedicle-screw-alone fixation and fusion to L5 was safe and effective in CMD patients with scoliosis of <95° and pelvic obliquity of <20°. Scoliosis curves were flexible (75% correction) on side-bending films preoperatively. Curve correction and maintenance of correction in the coronal and sagittal plane was excellent. The pelvic obliquity significantly improved. Balanced sitting posture was achieved and maintained in all patients. Our patients with CMD spinal deformity and a moderately and severely decreased FVC could be operated on safely and successfully with general anesthesia. All patients were extubated in the operating room. There were no major complications or deaths. We believe a FVC of <30% alone is not a predisposition to pulmonary complications. However, cardiomyopathy might be a determining risk of mortality, and we believe surgery for these patients should be avoided. Patients' and parents' satisfaction was high.

Offprint requests to: M. Takaso
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Introduction

Congenital muscular dystrophy (CMD), among the myopathic disorders, is one form of flaccid neuromuscular disorder (NMD). Patients with an NMD frequently develop progressive spinal deformities. In patients with an NMD, gradual deterioration is the hallmark. Motor strength and pulmonary function are key issues of quality of life (QOL) for patients with an NMD.[1] Spinal deformity is almost universal in these patients.[2,3] For NMD patients who have a severe spinal deformity, sitting is often difficult and is accompanied by pain and breakdown of the skin.[1–4] Progressive deterioration of respiratory muscle function is a major problem in these patients.[4–6] Moreover, their respiratory function can deteriorate owing to progression of the spinal deformity.[7–10]

Spinal deformity surgery in NMD patients has been aimed at maximizing function and improving patients' QOL.[1,4,8,9] This surgery has been highly effective in stabilizing the spine and maintaining upright, comfortable sitting balance.[1,3,6,8,11–16] Despite the magnitude of this surgery, most of the patients and their families have believed scoliosis surgery improves their function, cosmesis, sitting balance, and QOL.[1]

Spinal deformity surgery in these patients should be performed early enough in the course of the underlying neuromuscular disorder, when cardiac and pulmonary function is sufficient, so patients can be anesthetized and operated on relatively safely and to reduce the likelihood of major complications.[5,16,17] Many studies have reported significant rates of peri/postoperative complications in patients with NMDs.[18]

Although many studies have reported on the results of surgical treatment of neuromuscular spinal deformity, most previous research is in patients with Duchenne muscular dystrophy (DMD), cerebral palsy (CP), and spinal muscular atrophy (SMA). To our knowledge, there has been no study on the results of spinal deformity surgery in patients with CMD. The purpose of this study was to review the clinical and radiological results of spinal deformity surgery in a group of patients with CMD.

Materials and methods

The study was approved by the institutional review board (IRB) of Kitasato University, and informed consent was obtained from all patients prior to participation in the study. We obtained agreement for publication from the IRB of Kitasato University.

A total of 10 consecutive patients with a diagnosis of neuromuscular scoliosis secondary to CMD undergoing spinal deformity surgery between 2004 and 2009 were identified. A minimum 2-year follow-up was required for inclusion in this study. There were three patients with Fukuyama CMD, three with Ullrich CMD, and four with nonsyndromic CMD (merosin-negative). They were nonambulatory. All of the patients had standard posterior spinal fusion and pedicle-screw-alone fixation performed by the same surgeon (M.T.).

Our inclusion criteria required that each patient (1) had considerable difficulty with sitting balance, pain or breakdown of the skin due to scoliosis, and pelvic obliquity; (2) was able to ventilate his/her lung autonomously; (3) was not ventilator dependent; and (4) did not have cardiac failure. We usually recommend posterior spinal fusion and instrumentation to all CMD patients if the patient meets the inclusion criteria. Surgery was considered regardless of the severity of the spinal deformity and the contractures of the hips or knees, the patient's age, and the level of his or her intelligence. We are of the opinion that patients with difficulty sitting and/or who have pain or breakdown of the skin due to scoliosis can benefit from instrumentation and posterior spinal fusion for scoliosis and pelvic obliquity if the surgery is performed successfully. However, sufficient informed consent is important, and the decision to perform surgery must be made by the patient and his or her family with sufficient preoperative informed consent.

Cardiac function tests

Cardiac function was assessed by transthoracic echocardiography and electrocardiography preoperatively.

Pulmonary function tests

Pulmonary function tests were performed with a computerized pulmonary function system (FUDAC-50; Fukuda Denshi, Tokyo, Japan). The various pulmonary functions were assessed. The tests were performed on each occasion with the patients seated in a wheelchair. Arm-span measurements were selected for calculations of predicted heights. Pulmonary function value reported in this study is forced vital capacity (FVC) expressed as percent of the predicted value (%FVC) on the published normative data. Pulmonary function tests were evaluated in all the patients included in this study at 6 weeks before surgery, then each week, and the day before surgery. After surgery, pulmonary function tests were also evaluated at 6 weeks after surgery and then every 3 months thereafter. On each occasion, the %FVC was measured three times and evaluated by a single independent observer. The best trial for the test was used for further analysis. The initial 6-week follow-up period after surgery was chosen to avoid possible negative influences with this type of surgery (e.g., pain).