Risk factor analysis of kyphotic malalignment after cervical intramedullary tumor resection in adults

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OBJECTIVE A number of studies have reported that surgery for cervical intramedullary tumors via the posterior approach can result in postoperative sagittal malalignment of the cervical spine; however, the risk factors remain unclear. The purpose of this study was to investigate the changes in cervical spinal alignment after surgery for cervical intramedullary tumors in adults and to elucidate the risk factors for cervical spinal sagittal misalignment.

METHODS Data for the period from April 2001 to December 2011 for all adults who had undergone surgery for cervical intramedullary spinal cord tumors at a single institution were retrospectively analyzed to determine the postoperative changes in cervical spine alignment. Patients younger than 20 years of age and those who required postoperative radiotherapy were excluded from the study. Patients were divided into 2 groups according to tumor location: upper tumor (U) group, in which the central region of the tumor was above the C-5 level; and lower tumor (L) group, in which the central region of the tumor was at or below the C-5 level. Changes in alignment of the cervical spine were measured on plain lateral radiographs. Data on atrophy of the deep extensor muscles (DEMs), tumor location, detachment of the DEMs from the C-2 spinous process, the C2–7 angle before surgery, patient age at surgery, tumor histology, patient sex, tumor size, and number of laminae affected were reviewed for each patient, and the correlation of each of these factors with cervical spinal malalignment was evaluated using statistical analysis.

RESULTS The 54 adults eligible for analysis had a mean age of 49.1 years. Ependymoma was the most common cervical intramedullary tumor (63.0%) in this series. In the tumor location U group, the kyphotic angle of the C2–7 spinal segments increased after surgery ($-5.8^\circ \pm 2.8^\circ$). In contrast, in the L group, the C2–7 lordotic angle increased after surgery ($6.4^\circ \pm 2.6^\circ$). In the univariate analysis, atrophy of the DEMs, detachment of the DEMs from the C-2 spinous process, and an upper cervical location of the tumor were identified as factors significantly correlated with the development of cervical spinal kyphosis after surgery. Multiple linear regression analysis revealed the following as risk factors for kyphotic change of the cervical spine after surgery: 1) atrophy of the DEMs after surgery ($\beta = -0.54$, $p < 0.01$), and 2) detachment of the DEMs from the C-2 spinous process ($\beta = -0.37$, $p < 0.01$).

CONCLUSIONS Atrophy of the DEMs after surgery and detachment of the DEMs from the C-2 spinous process are directly related to the risk of cervical spinal kyphosis after surgery for cervical intramedullary tumors in adults. Therefore, preservation of the DEMs, especially those attached to the C-2 spinous process, is important for the prevention of kyphotic malalignment of the cervical spine after surgery for intramedullary tumors.

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KEY WORDS cervical intramedullary tumor; cervical alignment; kyphosis; deep extensor muscles; oncology

INTRAMEDULLARY spinal cord tumors are relatively rare neoplasms that account for 2%–4% of all central nervous system tumors.^{8,33} They are less common in adults than in children, accounting for 20% of all adult and 35% of all pediatric spinal tumors.^{3,5} With advances in

the microsurgical techniques for resection of these tumors, the management paradigm for these tumors has shifted to aggressive treatment with radical resection.^{7,8,23} With this approach, long-term survival and quality of life for patients have improved after surgery.^{7,8,23,33} Although current

ABBREVIATIONS CIST = cervical intramedullary spinal cord tumor; DEM = deep extensor muscle; ICC = intraclass correlation coefficient; JOA = Japanese Orthopaedic Association; L = lower tumor; U = upper tumor.

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treatment is aimed at radical or subtotal removal of these tumors, the postoperative development of progressive spinal deformities such as kyphosis is well recognized in children; such deformity has been reported to develop in 22%–100% of patients undergoing surgery for intradural spinal tumors.^{12,13,20,23,38} According to other studies, however, spinal deformity occurs at a rate of about 10% following surgery for intradural spinal tumors (including intramedullary tumors and extramedullary tumors) in adults.^{9,26} But no studies have examined the frequency of change in spinal alignment after surgery for intramedullary tumors in adults. Some pediatric studies have attempted to identify clinical predictors of spinal deformities after surgery for intramedullary tumors of mixed spinal regions, that is, tumors located in the cervical or thoracic spine level. But such studies may be inappropriate because the biomechanical properties of different spinal regions are different³⁸ and because the etiology of any deformities developing after surgery may differ among the various spinal regions. Furthermore, while a number of studies have reported on the development of cervical spinal deformities following surgery for cervical spinal cord tumors in adults,^{9,16,29} the tumors in these studies included intramedullary tumors and intradural extramedullary tumors, as well as extradural tumors. For intradural extramedullary tumors and extradural tumors, especially dumbbell tumors, there is a substantial risk of spinal deformities such as enlargement of the posterior arch and intervertebral foramina, as well as vertebral scalloping. In addition, the facet joint must sometimes be sacrificed for tumor resection. These factors confound any analysis of the pure effect of tumors inside the spinal cord because they can also affect the risk of cervical spine misalignment after surgery.

Since previous studies aimed at identifying risk factors for the development of misalignment of the spine have included tumors located in different spinal regions and patients of various age groups and with various pathologies, their validity and conclusions remain under debate. The purpose of the present study was to identify clinical predictors for the development of sagittal misalignment of the cervical spine following surgery for cervical intramedullary tumors in adults.

Methods

Subjects

Data for the period from April 2001 to December 2011 for all adults with cervical intramedullary spinal cord tumors (CISTs) who had undergone surgery at a single academic institution (Keio University Hospital) were retrospectively reviewed for the development of a cervical alignment change following treatment. Patients with cervicothoracic tumors (in which the central region of the tumor was the thoracic level), intradural extramedullary spinal cord tumors, and extradural spinal cord tumors were excluded. Patients younger than 20 years of age and those who required postoperative radiotherapy were also excluded. Patients were evaluated using the Japanese Orthopaedic Association (JOA) scoring system for cervical myelopathy both preoperatively and at 2 weeks postoperatively.

Radiological Assessment

Prior to surgery, plain lateral radiographs and MR images were obtained in all patients. Postoperatively, plain lateral radiographs and MR images were obtained at annually scheduled follow-up appointments to assess for the presence of cervical malalignment and tumor recurrence. According to the tumor location on the sagittal T2-weighted and gadolinium-enhanced MR images obtained before surgery, patients were divided into 2 groups: upper tumor (U) group, in which the central region of the tumor was above the C-5 level; and lower tumor (L) group, in which the central region of the tumor was at or below the C-5 level. Tumor size was defined as the longitudinal extent (number of vertebral columns) of tumors as assessed on the sagittal gadolinium-enhanced MR images. The sagittal T2-weighted MR images were also used for assessment of tumor size if the gadolinium-enhanced MR images were difficult to evaluate.

To identify changes in the curvature of the cervical spine, lateral cervical radiographs obtained at the final follow-up at least 1 year after surgery were compared with those obtained preoperatively. Curvature of the cervical spine was determined in the upright neutral position by measuring the tangential lines along the posterior borders of the C-2 and C-7 vertebral bodies, the C-2 and C-5 vertebral bodies, or the C-5 and C-7 vertebral bodies (C2–7 angle, C2–5 angle, and C5–7 angle, respectively). For a few patients who were unable to stand, we used the sitting neutral position. According to a previous study on alignment change after cervical spinal cord tumor resection, a C-2 laminectomy causes the greatest local kyphosis at the C4–5 level, whereas a C-7 laminectomy causes the greatest local lordosis at the C5–6 level,¹⁶ suggesting that the apex of the cervical sagittal alignment change after upper or lower laminectomies would be located at the C-5 level. Hence, we measured curvature of the cervical spine at C2–5 and C5–7 separately.

To quantify damage to the deep extensor muscles (DEMs), the muscle atrophy rate was calculated for each patient from the cross-sectional areas of the DEMs from C-2/C-3 to C-6/C-7 measured on the preoperative and postoperative (at the final follow-up at least 1 year after surgery) axial T2-weighted MR images obtained at 3 different intervertebral levels: the intervertebral level corresponding to the central region of the tumor and 1 level above and 1 below the level corresponding to the central region of the tumor. Analysis was achieved by manually tracing defined regions of interest within the fascial borders of the DEMs (semispinalis cervicis and multifidus muscles) bilaterally on the axial T2-weighted images (See Fig. 5D–I for an example, the area surrounded by the white line). The sum of these 3 cross-sectional areas was regarded as the total area for each patient. Cross-sectional areas of the DEMs were measured as previously described with slight modifications.^{6,21,25,30} The rate of muscle atrophy was calculated using the following formula, as previously described:³⁰ $(1 - \text{total postoperative area} / \text{total preoperative area}) \times 100$. A spine surgeon not directly involved in these surgeries (S.N.) measured the area of the DEMs at 3 different time points by using ImageJ software (Scion Corp., <http://rsb.info.nih.gov/ij/>). The average of the

3 measurements was used as the area of the DEMs. The intraobserver reliability tested by the intraclass correlation coefficient (1, 1) and ICC (1, 3) formula was 0.83 and 0.94, respectively. To assess the interobserver error, 60 randomly chosen axial T2-weighted MR images were measured independently by 3 spine surgeons not directly involved in these surgeries (S.N., A.I., and A.Y.). The interobserver reliability tested by ICC (2, 1) and ICC (2, 3) formula was 0.85 and 0.94, respectively.

Surgical Technique

For approaching the CISTs, osteoplastic laminoplasty spanning the length of the tumor was performed. For the osteoplastic laminoplasty, only the medial facet joint was exposed by subperiosteal paraspinal muscle dissection, and an effort was made to preserve the facet joint capsule in every case. No facetectomy was performed in any of the patients during surgery. We performed French-door laminoplasty according to the method described by Kurokawa,¹⁹ with some modifications. For tumorectomy from the C-2 to C-7 levels, after detaching the bilateral paravertebral muscles from the spinous processes, the laminae were split in the midline by oscillating the wire. Bilateral gutters were created as hinges at the borders of the laminae and facets. After the halves of the laminae were elevated, tumorectomy was performed. For the C-1 level tumorectomy, a C-1 laminectomy was performed. Ultrasonic aspiration was sometimes used to excavate tumor from the inside outward until its interface with the white matter was reached. Sensory evoked and motor evoked potentials were used for monitoring. Resection was deemed complete once the white matter interface was reached. At the end of the procedure, the split halves of the laminae were replaced and secured in place by anchoring sutures (Fig. 1). If the DEMs were detached from the C-2 spinous process during the procedure, reconstruction of extensor muscles was performed with suturing to the C-2 spinous process. No patient in this study had detachment of the semispinalis capitis muscles. Gross-total resection was at-

tempted in all cases. The same senior spine surgeons with expertise in spinal cord tumor surgery performed the operations in all cases.

Statistical Analysis

Statistical analyses were performed using the SPSS software (version 22, SPSS Inc.). A number of pre-, intra-, and postoperative variables were hypothesized to be associated with the risk of a C2–7 cervical alignment change after surgery for CIST. We analyzed these variables: atrophy of the DEMs, tumor location in the cervical spine, detachment of the DEMs from the C-2 spinous process, C2–7 angle before surgery, age at surgery, tumor histology, patient sex, tumor size, and number of laminae affected. Univariate analysis (correlation analysis) was conducted using all the variables except for tumor histology; the Kruskal-Wallis nonparametric test was used for the univariate analysis of tumor histology. Factors with $p < 0.25$ identified from the univariate analysis were entered into the multiple linear regression analysis. Differences were considered significant at $p < 0.05$.

Results

Patient Population

None of the patients required tumor resection combined with instrumented fixation. The study population of 54 patients included 38 males and 16 females, with a mean age of 49.1 ± 6.7 years (range 21–77 years) at the time of surgery. The mean duration of follow-up was 80.4 ± 10.9 months (range 30–144 months). None of the patients had any intrinsic bony lesions potentially affecting spinal alignment. The mean pre- and postoperative JOA scores were 12.5 ± 3.3 and 12.3 ± 3.6 , respectively. Clinical details for all patients are summarized in Table 1. There were 33 patients in the tumor location U group and 21 patients in the L group. The CISTs included hemangioma in 7 patients (13.0%), hemangioblastoma in 7 (13.0%), schwannoma in 1 (1.8%), ependymoma in 34 (63.0%),

FIG. 1. Surgical technique. French-door laminoplasty spanning the length of the tumor was performed for approaching the tumor. After detachment of the bilateral paravertebral muscles from the spinous processes, the centers of the laminae were cut (A). Bilateral gutters were created as hinges at the borders of the laminae and facets (B). After the halves of the laminae were elevated (C), tumorectomy was performed (D and E). The split halves of the laminae were replaced and secured in place by anchoring sutures at the end of the procedure (F). Copyright Satoshi Nori. Published with permission. Figure is available in color online only.

subependymoma in 1 (1.9%), and astrocytoma in 4 (7.4%). During the operations, detachment of the DEMs from the C-2 spinous process was performed in 26 patients (48.1%).

Cervical Alignment Change After Surgery

In the U group, kyphotic malalignment of the C2–7 spinal levels developed ($-5.8^\circ \pm 2.8^\circ$). In contrast, in the L group, an increase in the C2–7 lordotic angle was observed after surgery ($6.4^\circ \pm 2.6^\circ$; Figs. 2 and 3A). We found the same tendency for the C2–5 angle as for the C2–7 angle: We observed kyphosis of the C2–5 segment in the U group but lordosis of this spinal segment in the L group after surgery ($-5.7^\circ \pm 2.2^\circ$ and $7.1^\circ \pm 1.4^\circ$, respectively; Fig. 3B). On the other hand, we observed slight kyphotic malalignment of the C5–7 segment in the L group after surgery but almost no alignment change in the U group ($-2.3^\circ \pm 1.7^\circ$ and $-0.7^\circ \pm 1.3^\circ$, respectively; Fig. 3C).

Detachment of DEMs From C-2 Spinous Process and Development of Kyphotic Malalignment After Surgery

We also analyzed the relationship between detachment of the DEMs from the C-2 spinous process and cervical alignment change after surgery. Detachment of the DEMs from the C-2 spinous process (detached group) increased the rate of development of kyphosis at the C2–7 spinal levels after surgery ($-8.4^\circ \pm 3.1^\circ$), whereas in the patient group in which the DEMs were not detached from the C-2 spinous process (nondetached group), increased lordosis of the C2–7 spinal segment was noted ($6.3^\circ \pm 2.1^\circ$). We found the same tendency for the C2–5 angle as for the C2–7 angle: We observed kyphosis of the C2–5 segment in the detached group but lordosis of this spinal segment in the nondetached group after surgery ($-7.9^\circ \pm 2.3^\circ$ and $4.5^\circ \pm 4.7^\circ$, respectively). On the other hand, we observed slight kyphotic malalignment of the C5–7 segment in both groups after surgery ($-0.4^\circ \pm 1.5^\circ$ and $-2.1^\circ \pm 2.1^\circ$, respectively).

Risk Factors of Kyphotic Malalignment After Surgery

We performed statistical analyses to identify the risk factors for C2–7 malalignment after surgery. Univariate analyses revealed atrophy of the DEMs after surgery as being significantly correlated with kyphotic malalignment after surgery ($r = -0.67$, $p < 0.01$; Fig. 4). Furthermore, an upper tumor location as well as detachment of the DEMs from C-2 was significantly correlated with the risk of kyphotic malalignment after surgery ($r = -0.37$, $p < 0.01$; $r = -0.55$, $p < 0.01$, respectively). None of the other factors were statistically significantly related, as shown by the p values: C2–7 angle before surgery, $p = 0.11$; number of laminae affected, $p = 0.17$; tumor size, $p = 0.33$; age at surgery, $p = 0.88$; tumor histology, $p = 0.47$; patient sex, $p = 0.29$; preoperative JOA score, $p = 0.13$; postoperative JOA score, $p = 0.32$ (Table 2).

Next, we analyzed risk factors for the development of kyphosis after surgery by using multiple linear regression analysis. Interestingly, atrophy of the DEMs after surgery ($\beta = -0.56$, $p < 0.01$) and detachment of the DEMs from the C-2 spinous process ($\beta = -0.37$, $p < 0.01$) increased the risk of kyphotic malalignment after surgery. None of the

other examined factors were significantly associated with this risk (Table 3).

Illustrative Case

A 39-year-old man (Case 35) with an ependymoma belonging to the U group underwent C3–6 laminoplasty for gross-total resection (Fig. 5A). In this case, we detached the DEMs from the C-2 spinous process to secure the operative field. A preoperative lateral plain radiograph showed a lordotic angle of 0.4° of the C2–7 segment (Fig. 5B). However, 2 years 8 months after the surgery, kyphotic malalignment developed at C2–7 (-27.8° ; Fig. 5C). Preoperative and postoperative MRI showed atrophy of the DEMs (Fig. 5D–I) after surgery. The muscle atrophy rate in this patient was 36.7%.

Discussion

This is the first study focusing on clinical predictors for the development of sagittal misalignment of the cervical spine following surgery for CIST in adults. Here, we revealed that atrophy of the DEMs after surgery and detachment of the DEMs from the C-2 spinous process are directly related to the risk of developing cervical spinal kyphosis after CIST resection in adults.

A previous study showed that the development of spinal deformity after surgery for spinal cord tumors depended on the surgical level and the type of operation performed.¹⁶ Because we speculated that not only the surgical level but also the tumor level in the spine is associated with the risk of postoperative spinal deformity, we divided the patients into 2 groups according to tumor location in the cervical spine. Previously, most studies have been focused on the change in alignment of the entire C2–7 segment;¹⁷ thus, they often failed to detect local alignment changes or compensatory cervical alignment changes after surgery.¹⁶ By measuring the C2–7, C2–5, and C5–7 angles, we were able to evaluate both local and whole cervical alignment.

Kyphotic change at the C2–7 and C2–5 levels developed in the U group after surgery. In the L group, we observed lordotic change at the C2–7 and C2–5 levels after surgery. Moreover, detachment of the DEMs from the C-2 spinous process increased the C2–7 and C2–5 kyphotic angle after surgery, whereas C2–7 and C2–5 lordotic changes were observed in the group in which the DEMs were not detached. In a previous study of cervical spinal cord tumor patients who had undergone C-2 laminectomy, kyphosis in the upper cervical spine and a compensatory increase in lordosis of the lower cervical spine were observed. In contrast, in the patients who underwent C-7 laminectomy, local kyphosis developed in the lower cervical spine with a compensatory increase in lordosis of the upper cervical spine.¹⁶ Thus, the pattern of deformity depends on the laminectomy level.^{13,34} While we did not perform laminectomy, but rather double-door laminoplasty, we did obtain results consistent with those mentioned above. Although other studies have shown the preventive effect of laminoplasty against postoperative spinal deformity,^{16,23} our results indicated that laminoplasty can cause kyphotic change at the surgical level and compensatory lordotic change at other levels. No patient required deformity correction surgeries

TABLE 1. Clinical profile of patients who underwent surgery for CISTs

Case No.	Age at Surgery (yrs)	Sex	Pathology	Tumor Level	Laminoplasty or Laminectomy Level*	Detachment of DEM from C-2	FU (mos)	Preop C2–7 Angle (°)	Postop C2–7 Angle (°)
1	61	F	Hemangioma	U, C2–4	C1–4	Yes	86	–1.4	1.8
2	44	F	Hemangioma	U, C2–5	C1–5	Yes	81	6.5	10.1
3	44	M	Hemangioma	L, C-5	C4–6	No	67	2.2	2.4
4	38	M	Hemangioma	U, C-3	C2–4	Yes	48	16.8	14.5
5	47	M	Hemangioma	U, C3–4	C2–4	Yes	45	2.8	–1.5
6	30	M	Hemangioma	L, C6–7	C6–T1	No	33	7.8	19.3
7	71	F	Hemangioma	L, C6–7	C6–7	No	32	35.6	36.6
8	29	M	Hemangioblastoma	L, C-5	C4–6	No	128	13	9.3
9	33	M	Hemangioblastoma	U, C3–5	C3–7	No	115	2	15.6
10	37	M	Hemangioblastoma	U, C4–5	C3–5	No	92	1.7	6.3
11	49	F	Hemangioblastoma	U, C4–5	C4–6	No	80	2	14.4
12	45	M	Hemangioblastoma	U, C2–4	C1–4	Yes	74	3.8	2.2
13	32	M	Hemangioblastoma	U, C1–2	C1–3	Yes	62	10.1	–5.4
14	28	M	Hemangioblastoma	U, C2–5	C2–5	Yes	57	43.8	14.3
15	61	M	Neurinoma	U, C2–3	C1–3	Yes	66	34.9	32.5
16	34	M	Ependymoma	U, C3–4	C3–T2	No	144	–2.5	25
17	52	M	Ependymoma	U, C2–3	C1–4	Yes	140	22	16
18	24	F	Ependymoma	L, C-5	C4–7	No	127	–4	–15
19	68	M	Ependymoma	U, C2–4	C1–5	Yes	120	26	21.1
20	64	M	Ependymoma	L, C4–7	C3–T1	No	119	0	20.6
21	65	M	Ependymoma	L, C5–7	C4–T1	No	110	18	36.2
22	41	F	Ependymoma	U, C3–4	C2–T1	Yes	109	4	4.3
23	62	M	Ependymoma	U, C3–4	C1–6	Yes	103	33	35.6
24	38	M	Ependymoma	U, C0–3	C1–3	Yes	102	14	–5.5
25	54	M	Ependymoma	L, C6–7	C4–T2	No	100	1.9	19.1
26	35	F	Ependymoma	U, C4–5	C3–6	No	97	10.7	10.3
27	36	F	Ependymoma	U, C2–6	C1–T1	Yes	95	–9.5	–50.5
28	21	M	Ependymoma	U, C2–3	C2–T1	Yes	91	11.4	30.3
29	57	M	Ependymoma	L, C5–6	C3–6	No	88	16.8	19.6
30	29	M	Ependymoma	L, C6–T1	C4–T1	No	85	12.7	23.4
31	59	F	Ependymoma	L, C6–7	C4–7	No	82	3.9	4.7
32	45	M	Ependymoma	L, C5–6	C4–7	No	82	11.6	6.9
33	66	F	Ependymoma	U, C1–2	C1–4	Yes	78	19.9	12.8
34	65	M	Ependymoma	U, C1–2	C1–6	Yes	77	31.5	39.3
35	39	M	Ependymoma	U, C3–5	C3–6	Yes	77	0.4	–27.8
36	59	M	Ependymoma	U, C-2	C2–3	Yes	76	18.3	1.9
37	65	M	Ependymoma	U, C3–4	C3–5	No	75	8.4	18.7
38	59	F	Ependymoma	U, C2–3	C2–4	Yes	73	24.5	16.2
39	77	F	Ependymoma	U, C3–4	C2–5	Yes	72	30.7	17.7
40	65	M	Ependymoma	L, C5–6	C4–6	No	72	3.2	7.7
41	41	M	Ependymoma	L, C4–6	C3–7	No	71	–1	17.5
42	32	M	Ependymoma	L, C5–6	C5–7	No	63	5.5	0.2
43	63	F	Ependymoma	U, C1–2	C1–3	Yes	63	27.8	4.7
44	58	F	Ependymoma	U, C-4	C3–5	No	58	20.9	18.1
45	57	M	Ependymoma	U, C-2	C1–4	Yes	57	20.1	3.3
46	66	M	Ependymoma	L, C5–6	C4–6	No	51	26.8	13.1
47	36	M	Ependymoma	L, C5–T1	C4–T2	No	46	20	27.4
48	51	M	Ependymoma	U, C2–6	C1–7	Yes	41	17.4	–29.8
49	39	M	Ependymoma	U, C1–2	C1–3	Yes	36	17.7	13.7

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TABLE 1. Clinical profile of patients who underwent surgery for CISTs

Case No.	Age at Surgery (yrs)	Sex	Pathology	Tumor Level	Laminoplasty or Laminectomy Level*	Detachment of DEM from C-2	FU (mos)	Preop C2–7 Angle (°)	Postop C2–7 Angle (°)
50	60	F	Subependymoma	L, C5–7	C5–T1	No	30	21.4	46.3
51	63	M	Astrocytoma	L, C6–7	C5–T2	No	121	11	23.7
52	58	M	Astrocytoma	U, C2–3	C1–4	Yes	88	36.1	35.7
53	49	F	Astrocytoma	L, C4–7	C4–T1	No	87	14.6	8.9
54	49	M	Astrocytoma	L, C6–7	C5–7	No	70	7.2	34.6

FU = follow-up.

* Laminectomy was performed only at C-1.

for cervical malalignment after tumor resection during the study period. However, we think that paresis, dysphagia, and unbearable pain due to the developing cervical malalignment following tumorectomy would be indications for those surgeries.

Next, we analyzed the risk factors for a C2–7 alignment change after surgery using univariate and multivariate analyses. These analyses identified 2 main factors as influencing the risk of kyphotic change after tumor resection: 1) atrophy of the DEMs, and 2) detachment of the DEMs from the C-2 spinous process. Notably, atrophy of the DEMs after surgery was found to be significantly as-

sociated with the risk of postoperative kyphotic alignment change. From a biomechanical standpoint, the extensor musculature forces on the cervical spine were analyzed in a previous study, and its authors hypothesized that the semispinalis cervicis and semispinalis capitis muscles were primarily responsible for extension of the cervical spine and head.²⁴ Disruption of the extensor musculature caused by muscle detachment during surgery could promote sagittal plane deformity. In another study performed using a 3D computerized biomechanical model, investigators showed that the semispinalis cervicis and semispinalis capitis muscles generate 37% of the total strength

FIG. 2. Representative pre- and postoperative images from the U and L tumor location groups. Case 45. **A:** Preoperative sagittal gadolinium-enhanced MR image obtained in a 57-year-old man with an ependymoma belonging to the U group who underwent C-1 laminectomy and C2–4 laminoplasty for tumorectomy. **B:** Preoperative lateral plain radiograph. **C:** Lateral plain radiograph obtained 2 years 3 months after surgery, showing an increase in the C2–7 kyphotic angle. Case 6. **D:** Preoperative sagittal gadolinium-enhanced MR image obtained in a 30-year-old man with a hemangioma belonging to the L group who underwent C6–T1 laminoplasty for tumorectomy. **E:** Preoperative lateral plain radiograph. **F:** Lateral plain radiograph obtained 1 year 6 months after surgery, showing an increase in the C2–7 lordotic angle.

FIG. 3. Cervical alignment change after surgery. **A:** Preoperative and postoperative C2–7 alignment change in the U and L tumor location groups. In the U group, the C2–7 kyphotic angle increased postoperatively ($-5.8^\circ \pm 2.8^\circ$). In the L group, the C2–7 lordotic angle increased after surgery ($6.4^\circ \pm 2.6^\circ$). **B:** Postoperative change in the C2–5 spinal alignment in the U and L groups. In the U group, the C2–7 kyphotic angle increased postoperatively ($-5.2^\circ \pm 2.2^\circ$). In the L group, the C2–5 lordotic angle increased after surgery ($7.1^\circ \pm 1.4^\circ$). **C:** Preoperative and postoperative C5–7 alignment change in the U and L groups. While there was almost no change in alignment in the U group ($-0.7^\circ \pm 1.3^\circ$), slight cervical kyphosis was observed in the L group ($-2.3^\circ \pm 1.7^\circ$).

produced by all the neck muscles in isometric neck extension.³⁶ Moreover, a previous cadaveric study showed that cervical instability is primarily caused by detachment of the semispinalis cervicis muscle from the C-2 spinous process.¹ The C-2 vertebral level has the largest spinous process, which serves as the site of attachment for more extensor muscles than any other cervical vertebral spinous process. It plays a principal role as a lever arm.³² In one study that analyzed cervical instability after laminectomy for spinal cord tumor resection, the C-2 laminectomy was found to be directly related to the development of cervical instability; this might be explained by the fact that C-2 laminectomy causes a reduction in the area of insertion of the semispinalis cervicis muscle.¹⁷ Therefore, several authors have advocated laminoplasty with reattachment of the removed muscles to the C-2 spinous process or laminoplasty with a DEM-preserving procedure for all patients who require C-2 decompression.^{15,16,22,24,32} In the present study, although we reattached the DEMs to the C-2 spinous process, postoperative kyphotic malalignment was still observed. Hence, once the DEMs are detached from the C-2 spinous process, reattachment of the muscles may not be effective in preventing postoperative cervical malalignment. This is consistent with a previous study comparing the cervical alignment change between open-door

laminoplasty involving elevation of the C-3 lamina with reattachment of the DEMs to the C-2 spinous process and open-door laminoplasty with a C-3 dome laminotomy or laminectomy to completely preserve the insertion of the DEMs to C-2.¹⁴

Our statistical analyses identified tumor location, C2–7 angle before surgery, tumor size, tumor histology, age at surgery, patient sex, and number of laminae affected as not significantly associated with the risk of alignment change of the cervical spine following surgery. Previous studies have shown that a preexisting kyphotic deformity tends to increase the risk of malalignment of the cervical spine after surgery.^{17,38} In the present study, however, we found no significant relationship between the C2–7 angle before surgery and the risk of postoperative cervical spinal malalignment. This difference could perhaps be explained by the low incidence of preoperative cervical malalignment in our patients; only 5 of the 54 patients showed kyphotic malalignment of the cervical spine before surgery, and the maximum kyphotic angle measured preoperatively was only -9.5° . Therefore, we believe that the absence of any remarkable kyphotic deformity before surgery may explain why the preoperative C2–7 angle was not identified as a risk factor for cervical spinal malalignment after surgery.

Spinal deformities in children can be caused by spinal cord lesions alone, even in the absence of surgical interven-

FIG. 4. Correlation between the risk of cervical spinal misalignment and atrophy of the DEMs. Analysis revealed that atrophy of the DEMs at the tumor level was strongly correlated with the development of cervical kyphosis after surgery ($r = -0.67$, $p < 0.01$).

TABLE 2. Univariate correlation analyses of C2–7 alignment

Characteristic	r	p Value
Atrophy of DEMs	-0.67	8.42E-08
Detachment of DEMs from C-2	-0.55	1.60E-05
Upper localization of tumor	-0.37	6.30E-03
C2–7 angle before surgery	-0.22	0.11
No. of laminae affected	0.19	0.17
Size of tumors	0.14	0.33
Age at surgery	0.02	0.88
Sex	0.15	0.29
Preop JOA score	-0.22	0.13
Postop JOA score	-0.15	0.32

TABLE 3. Multiple linear regression analyses of C2–7 kyphotic change

Characteristic	β	p Value
Atrophy of DEMs	-0.56	2.00E-06
Detachment of DEMs from C-2	-0.37	7.35E-04
Upper localization of tumor		0.43
C2–7 angle before surgery		0.53
No. of laminae affected		0.42
Preop JOA score		0.17

tion.^{20,35} Some previous studies have speculated that compression of the motor neurons in the anterior horn causes cervical muscle denervation and weakness, which can lead to cervical spinal deformities.^{10,34} However, even though gross-total resection was attempted in all patients in the present study, there was no correlation between tumor size and cervical alignment change. This discrepancy may be attributable to anatomical differences between children and adults such as the horizontal cervical spine facets, the lax ligamentous structures, and the growing vertebral column in children. It has been reported that a young age is one of the risk factors for cervical spinal deformities after surgery for spinal tumors. Pediatric patients are known to be at the greatest risk for developing deformities, followed

FIG. 5. Case 35. **A:** Preoperative sagittal T2-weighted MR image obtained in a 39-year-old man with an ependymoma belonging to the U group who underwent C3–6 laminoplasty for gross-total resection of the tumor. **B:** Preoperative lateral plain radiograph. **C:** Lateral plain radiograph obtained 2 years 8 months after surgery, showing an increase in the C2–7 kyphotic angle. **D–F:** Preoperative axial T2-weighted MR images obtained at the C2–3, C3–4, and C4–5 intervertebral levels. **G–I:** Post-operative images obtained at the same vertebral levels. *White lines* show the areas of measurement of the DEMs. Muscle atrophy of the DEMs was observed after surgery.

by young adults (< 25 years old), who also have an elevated risk as compared with that in older adults.^{26,37} Since our study was focused on adults with CIST, only 1 young adult patient and no pediatric patients were included in this study. This could explain the absence of any significant correlation between age and the risk of cervical spinal alignment change observed in our study.

While some studies have shown no correlation between the number of laminae affected and the incidence of cervical deformity,^{4,37} another study has reported that the number of laminae affects the risk of cervical spinal malalignment after spinal surgery.¹⁷ Note, however, that tumors in the latter study included not only intramedullary tumors but also intradural extramedullary tumors and extradural tumors. Hence, there is a substantial risk of spinal deformities due to the bone instability caused by enlarging tumors as well as by sacrificing the facet joint during tumorectomy. These factors confound any analysis of the pure effect of the number of laminae affected and cause the discrepancy in our results that the number of laminae affected did not correlate with cervical alignment change.

Another possible risk factor that has been associated with a cervical alignment change after surgery for CIST in adults is the extent of destruction of the facet joints. Biomechanically, spinal instability increases after laminectomy when at least 50% of the facet joints are removed.^{27,39} It has also been reported that cervical instability is most frequent and most severe when the facet joints are destroyed by tumor or surgery.¹⁷ Since our series only included intramedullary tumors, we did not have to sacrifice the facet joints in any of our cases during surgery, nor were the facet joints destroyed by tumor. Therefore, we excluded destruction of the facet joints as a variable from our risk factor analyses. Radiation therapy has also been documented as a risk factor, both experimentally and clinically, for the development of spinal deformity in children and adults. The reported incidence of spinal deformity after radiation therapy in previous studies is about 60%.^{2,11,18,28} Since we excluded from our series those patients who had undergone radiation therapy, we also eliminated this factor from our risk factor analysis.

Study Limitations

This study has several limitations. The major limitation is its inherent nature as a retrospective study. Another limitation is the small sample size. Given the lack of long-term surgical outcome data in most patients, it was difficult to evaluate the effect of postoperative cervical malalignment on long-term postoperative functional and quality-of-life outcomes.

Conclusions

Following surgery for CIST in adults, kyphotic change at the surgical level and compensatory lordotic change at other levels were often observed. Statistical analyses to identify factors that might be related to the C2–7 alignment change observed after surgery revealed that atrophy of the DEMs after surgery and detachment of the DEMs from the C-2 spinous process are directly related to the risk of developing cervical kyphosis. On the other hand,

tumor location, C2–7 angle before surgery, tumor size, tumor histology, age at surgery, patient sex, or number of laminae affected were not significantly associated with cervical malalignment after surgery for CIST in adults.

To avoid cervical spinal malalignment after surgery for CIST, we suggest considering preservation of the DEMs (especially those attached to the C-2 spinous process) during the tumor resection, such as by muscle-preserving posterior arch recapping.³¹ Alternatively, posterior fusion might be considered for the patients whose DEMs are detached from the C-2 spinous process.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Iwanami, Nori, Nakamura. Acquisition of data: Iwanami, Nori, Yasuda, Momoshima. Analysis and interpretation of data: Nori. Drafting the article: Iwanami, Nori, Nagoshi, Nakamura. Critically revising the article: Iwanami, Nori, Nagoshi. Reviewed submitted version of manuscript: Iwanami, Nori, Yasuda, Nagoshi, Fujita, Hikata, Yagi, Tsuji, Watanabe, Matsumoto, Nakamura, Ishii. Statistical analysis: Nori. Study supervision: Iwanami, Yasuda, Nagoshi, Fujita, Hikata, Yagi, Tsuji, Watanabe, Matsumoto, Nakamura, Ishii.

Supplemental Information

Previous Presentations

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