Risk factors for a poor outcome following surgical treatment of cervical spondylotic amyotrophy: a multicenter study

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Abstract

Introduction Cervical spondylotic amyotrophy (CSA) is characterized by muscle atrophy in the upper extremities without gait disturbance. However, the indications and outcomes of surgical treatment for CSA have not been clarified. The purpose of this study was to determine the risk factors for a poor outcome following surgical treatment of CSA.

Materials and methods We performed a retrospective review of CSA in patients from 1991 to 2010 through a multicenter study. We collected information regarding age, type of muscle atrophy, preoperative manual muscle test (MMT), duration of symptoms, high-intensity areas on T2-weighted MR images, low-intensity areas on T1-weighted MR images, levels of spinal canal stenosis, cervical kyphosis and surgical procedures (laminoplasty, anterior

cervical discectomy and fusion and posterior spinal fusion), and calculated overall risk factors related to a poor outcome following surgery. Univariate analyses and multivariate logistic regression analysis were performed to identify correlates of a poor outcome.

Results Fifty-nine patients, 95 % male (56 patients), were included in our analysis with a mean age of 59 years (range 32–78 years). Eighteen patients did not improve after surgery. Symptom duration (OR = 1.263), preoperative MMT grade (OR = 0.169) and distal type of CSA (OR = 9.223) were all associated with an increased risk of a poor surgical outcome.

Conclusion Early surgery is recommended for CSA patients in whom conservative treatment has not been successful. We also recommend surgery for patients who have severe preoperative muscle weakness or have the distal type of CSA.

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Keywords Cervical spondylotic amyotrophy · Poor outcome · Risk factor · Surgical treatment

Abbreviations

CSA	Cervical spondylotic amyotrophy
CSM	Cervical spondylotic myelopathy
ALS	Amyotrophic lateral sclerosis
MMT	Manual muscle test
HIA	High-intensity areas
LIA	Low-intensity areas
ACDF	Anterior cervical discectomy and fusion
PSF	Posterior spinal fusion
OR	Odds ratio
EMG	Electromyography

Introduction

In 1952, Brain et al. [1] first reported on a case of cervical spondylosis with muscle atrophy of the upper extremities but without sensory disturbance. Keegan [2] later reported on this case at autopsy and described this condition as dissociated motor loss in the upper extremities with cervical spondylosis. In Japan, Yanagi et al. [3] labeled this condition “Cervical Spondylotic Amyotrophy (CSA)”. After analyzing the clinical features of CSA, they concluded that this condition was caused, not only by mechanical compression of the spinal cord, but also by selective damage to the anterior horn of the spinal cord secondary to circulatory disturbance. Kameyama et al. [4] reported that the pathophysiological basis for this syndrome was multisegmental damage possibly caused by circulatory insufficiency.

Cervical spondylotic amyotrophy is classified as either a proximal or distal type of muscle atrophy. The proximal type of CSA is characterized as weakness of the deltoid and biceps, such as a drop shoulder, whereas, the distal type is characterized as weakness of the hand muscles such as a drop finger. CSA patients are in their 30s–70s, but most are over 50 years old.

Usually, patients with CSA are first treated conservatively, such as with cervical traction or a neck collar. If the conservative treatment is ineffective, surgery is performed. Most cases have been successfully treated with surgery [5–7]; however, some surgeries have resulted in poor outcomes. It is in the best interest of patients and surgeons to understand potential risk factors related to a poor outcome following surgery. Several studies on CSA mentioned risk factors related to a poor outcome following surgery [6, 7], but there has not been a full multivariate analysis on this

type of data. To the best of our knowledge, this is the first report incorporating a multivariate analysis on the results of surgical treatment for CSA. Our goal was to conduct a retrospective case–control study and multivariate analysis to assess the risk factors for a poor outcome after surgical treatment for CSA.

Materials and methods

All institutions that participate in the Nagoya Spine Group (NSG) obtained institutional review board approval for this study. We performed a retrospective review on both prospectively and retrospectively collected data for this study. The medical records of patients who underwent surgical treatment from January 1991 to December 2010 were reviewed to identify those who did not improve after CSA surgery. Orthopedic spine surgeons performed all the surgical procedures on 59 patients with CSA at hospitals in the NSG. Ten institutions participated in this multicenter study, and 18 surgeons performed the operations on CSA patients. We collected data on patients’ ages, types of muscle atrophy, preoperative manual muscle test (MMT) results, duration of symptoms, high-intensity areas (HIA) on T2-weighted MR images (both sagittal and axial views), low-intensity areas (LIA) on T1-weighted MR images, levels of spinal canal stenosis, presence of cervical kyphosis and surgical procedures [laminoplasty, anterior cervical discectomy and fusion (ACDF) and posterior spinal fusion (PSF)]. We examined those factors related to poor outcomes following CSA surgery by applying univariate analyses and multiple logistic regression analysis of the surgical outcomes. To evaluate the effect of surgical treatment, we used MMT, and improvements in the muscle strength of the most atrophic and impaired muscles were classified into 4 grades: “excellent”, full recovery or recovery to a MMT grade of 2; “good”, 1 grade of recovery; “fair”, no improvement; “poor”, worsening effect. Taken together, we categorized two groups: a good outcome included “excellent” and “good”; a poor outcome included “fair” and “poor”. We performed statistical analyses between the two groups.

To differentiate between amyotrophic lateral sclerosis (ALS) and CSA, we always examined for the presence of sensory disturbance, bulbar symptoms and diffuse muscle atrophy, and also performed electromyography (EMG) in some cases before surgery. Furthermore, a neurologist ascertained that these patients did not have motor neuron disease before surgery. Finally, there were no cases which were diagnosed as ALS or other motor neuron disease after surgery.

Data analysis

Data were analyzed using SPSS (version 18.0). Univariate analyses were performed to examine the relationship between outcome at the final follow-up and prognostic factors. We used Mann–Whitney *U* tests for non-normally distributed variables, and Chi-square tests for categorical variables. Variables were included in a logistic regression model if their univariate analysis *p* value was ≤ 0.15 . The threshold for significance was a *p* value of <0.05 .

Results

A total of 59 patients with CSA underwent surgical treatment. There were 56 men and 3 women, with an average age of 59.4 years (range 32–78 years). The mean follow-up period was 2.7 years (range 1–12 years and 9 months). The duration of symptoms averaged 11.4 months (range 0.8 months to 15 years). Forty-one patients had proximal-type CSA, and 18 patients had distal-type CSA. HIA on T2 MRI was confirmed in 21 of 59 patients, and LIA on T1 MRI was confirmed in five of 59 patients. Spinal canal stenoses were found at an average of 2.7 intervertebral levels (range 1–5 levels). Forty-five patients received laminoplasty with or without foraminotomy, eight patients received ACDF, and six patients received PSF with laminoplasty/laminectomy. The surgical results were excellent for 33 patients, good for 8, fair for 17, and poor for 1 (Table 1). On univariate analyses of surgical outcomes, the duration of symptoms and preoperative MMT grade were statistically associated with a poor outcome after surgery ($p < 0.05$). There was a trend for patients with the distal type of muscle atrophy to have a poorer surgical outcome compared to patients with the proximal type of muscle atrophy ($p = 0.123$). Also, there was a trend in patients who received ACDF to have a better surgical outcome compared to patients receiving other surgical procedures ($p = 0.09$) (Table 2). As described in the “Materials and methods” section, potentially significant predictors of a poor outcome were used to fit a logistic regression model. On multivariate analyses of surgical outcomes, preoperative MMT grade and distal-type CSA were significant predictors of poor surgical outcome ($p \leq 0.05$) (preoperative MMT grade: OR = 0.169, and distal type of CSA: OR = 9.223), whereas, symptom duration was a highly significant predictor (OR = 1.263, $p \leq 0.001$). However, type of surgical procedure was not significantly associated with a poor surgical outcome. The full model is presented in Table 3.

Table 1 Demographic data of CSA patients

	All patients (<i>n</i> = 59)
Mean age (years)	59.4 (32–78)
Male/female	56/3
Mean follow-up (years)	2.7
Duration of symptoms (months)	11.4
Type of muscle atrophy	
Proximal	41
Distal	18
Preoperative MMT grade	2.3
Levels of stenosis	2.7
HIA on T2 MRI	21
LIA on T1 MRI	5
Kyphosis	17
Surgical procedure	
Laminoplasty	45
ACDF	8
PSF + laminoplasty/laminectomy	6
Surgical outcome	
Excellent	33
Good	8
Fair	17
Poor	1

CSA cervical spondylotic amyotrophy, MMT manual muscle test, PSF posterior spinal fusion, ACDF anterior cervical discectomy and fusion, HIA high-intensity area, LIA low-intensity area

Discussion

In our study, we demonstrated clinical features of CSA and used a logistic regression model to determine the risk factors related to having a poor postoperative outcome. We demonstrated that patients with poor surgical outcomes tended to have CSA symptoms for a longer duration and to have lower preoperative MMT grades than those patients with good outcomes (OR: 1.263, $p = 0.006$; OR: 0.169, $p = 0.015$, respectively). Based on our results, we recommend surgery for CSA patients with MMT grades of 1 or 2, and if conservative treatment has not been successful for 3–6 months. Furthermore, in our study, the surgical outcome for distal-type CSA was inferior to the outcome for proximal-type CSA (OR, 9.223; $p = 0.025$). Similarly, Uchida et al. [7] reported that surgical outcome in patients with the distal type of muscle atrophy was inferior to that in patients with the proximal type. Neuroradiologically, Kaneko et al. [8] mentioned that the pathophysiology of distal-type CSA included widespread gray matter lesions with less involvement of the lateral posterior spinal column. Fujiwara et al. reported that muscle power improved in 92 % of the proximal-type cases but only

Table 2 Univariate analyses of surgical outcome in 59 patients with CSA

	Group 1 Good (<i>n</i> = 41)	Group 2 Poor (<i>n</i> = 18)	<i>p</i> value
Age	58.9	60.4	0.57
Duration of symptoms (months)	4.9	27.3	<0.0001
Preoperative MMT grade	2.5	1.9	0.019
Type of muscle atrophy			
Proximal	31	10	
Distal	10	8	0.123
Levels of stenosis	2.7	2.9	0.517
HIA on T2 MRI	16	5	0.406
LIA on T1 MRI	4	1	0.514
Kyphosis ≥ 10	7	5	0.272
Surgical procedure			0.09
Laminoplasty	30	15	
ACDF	8	0	
PSF	3	3	

Table 3 Multiple logistic regression analysis for the risk factors of poor postoperative outcome

Variable	OR	<i>p</i> value	95 % CI
Duration of symptoms (months)	1.263	0.006	1.070–1.492
Preoperative MMT grade	0.169	0.015	0.040–0.710
Distal-type CSA	9.223	0.025	1.329–63.979
Surgical procedure	1.431	0.970	0.084–24.376

CI confidence interval

improved in 38 % of the distal-type cases. They speculated that the distal type basically involves impingement against the anterior horn which has less ability than the ventral nerve roots to regenerate [5].

In the present study, the HIA on T2 MRI, the LIA on T1 MRI, the number of vertebral levels with cervical canal stenosis, and the presence of cervical kyphosis were not significantly associated with a poor postoperative outcome. It has been reported that edema, myelomalacia and gliosis involve high-signal intensity on spinal cord T2-weighted MR images and low-signal intensity on T1-weighted MR images, suggesting irreversible changes of the spinal cord, and this signal intensity change was significantly associated with the poor postoperative outcome of cervical spondylotic myelopathy (CSM) [9–12]. In CSM patients, the intramedullary signal intensity change is usually consistent with damage in the central portion of the spinal cord [9]. However, CSA occurs with compression and damage of the anterior horn of the spinal cord or the ventral nerve root. This may be why there is no correlation between

signal intensity change on MRI and a poor surgical outcome. In fact, in our series, all cases which showed the signal intensity change demonstrated damage to the central portion of the spinal cord.

Previous reports also suggested that cervical kyphosis ≥ 10 was associated with a poor surgical outcome in CSM patients [13, 14]. Although our results indicated that cervical kyphosis was not associated with a poor surgical outcome, severe kyphosis may still influence outcome when laminoplasty is performed because kyphosis limits appropriate posterior shifting of the cord from the anterior compression. However, when root impingement, not compression of the anterior horn of the cord causes the muscle atrophy in CSA patients, cervical kyphosis would not be associated with surgical outcome.

On the other hand, Inui et al. [6] reported on conservative and surgical treatments for CSA patients. They mentioned that patients who fulfilled predictive factors such as age <50 years, duration of symptoms <6 months, single-level stenosis, foraminal stenosis, and a good response to traction therapy improved with conservative treatment. Although they attempted to determine the predictive factors related to surgical outcome, there was no statistical difference between the improvement group and the no improvement group.

Both ALS and CSA can have similar symptoms, and it can be difficult to distinguish the clinical manifestations of CSA during early stages of the disease [15]. Furthermore, although the clinical presentation of ALS closely resembles CSA, and both diseases preferentially affect middle-aged and elderly populations [16], it is very important to understand the differences between the diseases in order to distinguish one from the other prior to surgery. Cervical spondylosis does not cause atrophy of the tongue or fasciculations as we see with ALS. Amyotrophic lateral sclerosis patients can also have bulbar muscle involvement, articulation disorder and dysphagia. Muscle atrophy around the neck, such as the sternocleidomastoid muscle, and diffuse muscle atrophy of upper extremities are affected in ALS patients, but CSA involves only a few muscles. Apparent segmental sensory disturbance is characteristic of cervical spondylosis but not ALS, and ALS is more likely when there are widespread denervation potentials on EMG. However, if ALS cannot be definitively ruled out, surgery should not be performed, and patients should be carefully observed because ALS is always progressive.

The appropriate surgical methods for CSA are controversial. Some authors advocated that anterior decompression is reasonable for eliminating the anterior and anterolateral lesions [17–20]. The results from our series demonstrated a tendency for a better surgical outcome with ACDF than the other procedures, but it was not significant on univariate analysis ($p = 0.09$). Srinivasa et al. [19]

Fig. 1 **a** Sagittal image on T2 MRI showing multiple stenoses of the cervical spine. **b** Axial image on T2 MRI showing compression of the right anterior horn of the spinal cord (C4/5) and nerve root

reported that central corpectomy resulted in a good and long-lasting surgical outcome for distal-type CSA patients. However, these were elderly CSA patients with a tendency to have multiple and foraminal stenoses due to spondylosis making it difficult to correctly identify the lesion affecting the spinal cord or nerve root (Fig. 1). Thus, when patients had more than three lesions, we performed laminoplasty, whereas, with 1–2 levels of stenosis we performed ACDF. If the patients had severe kyphosis, we performed PSF with laminoplasty/laminectomy. In the present study, laminoplasty, with or without foraminotomy, was the most popular and effective operation for these patients. Our results demonstrated good outcomes for 30 of the 45 patients (67 %) treated by laminoplasty. We decided to add

foraminotomy in case foraminal stenosis was causing the root impingement. Previously, some authors also reported laminoplasty and foraminotomy as effective treatments for most patients with this syndrome [5, 20].

Several limitations of our study must be acknowledged. First, this was a retrospective, observational survey, and there was no standardized treatment protocol due to the number of surgeons at different hospitals involved in the care of these patients. Second, since this was a multicenter study, different surgeons have different skill levels which might influence the surgical outcome. Third, our patient sample size is small; it was underpowered for statistically evaluating the difference in risk for a poor outcome following surgery. However, this study is the first report in which we identify the risk factors related to a poor outcome through multivariate analysis, and this series contained the largest number of surgically treated CSA patients so far reported.

In conclusion, the present study offers a foundation for a deeper understanding of the factors that influence the risks of a poor outcome following surgical treatment in CSA patients. Early surgery is recommended for CSA patients in whom conservative treatment has not been successful and the clinical status before surgery (symptom duration, pre-operative MMT grade and distal type of CSA) significantly influences the surgical outcome. This information may be useful for clinicians in counseling patients and family members, and to develop more realistic treatment expectations for CSA patients.

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

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