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Clinical Studies

Comparison of fusion versus non-fusion surgery for retro-odontoid pseudotumor with atlanto-axial subluxation

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ABSTRACT

Background: Due to the limited number of reports comparing posterior fusion with posterior decompression alone for retro-odontoid pseudotumor, there remains no consensus on treatment preference, especially in older patients. This study compared posterior fusion (with or without additional decompression) with posterior decompression alone for treating spinal cord pressure from non-inflammatory retro-odontoid pseudotumor with atlanto-axial subluxation (AAS).

Methods: Forty-one patients (27 male and 14 female; mean age, 73.0 ± 11.4 years) who underwent either posterior cervical fusion or decompression alone for the treatment of non-inflammatory retro-odontoid pseudotumor with AAS and were observed for more than 1 year between September 2009 and July 2019 were enrolled. Thirty-two patients (23 male and 9 female; mean age: 71.8 ± 10.9 years) received posterior fusion surgery (fusion group) and 9 patients (4 male and 5 female; mean age: 77.2 ± 12.5 years) underwent decompression alone (non-fusion group). We compared pre- and postoperative Japanese Orthopaedic Association (JOA) scores and preoperative cervical alignment parameters between the groups.

Results: In the fusion group, the mean preoperative JOA score was significantly improved from 9.0 ± 3.2 points to 11.7 ± 3.2 points at the final follow-up ($p = 0.0002$). Similarly in the non-fusion group, the mean preoperative and final follow-up JOA scores were 8.2 ± 3.5 points and 11.7 ± 3.8 points, respectively ($p = 0.003$). The recovery rate at the final follow-up was 22.6% in the fusion group and 43.4% in the non-fusion group, which were statistically comparable ($p = 0.23$). We observed no remarkable correlations between cervical sagittal spinal alignment parameters and JOA score recovery rate in the cohort, nor was any significant subluxation progression seen.

Conclusion: Compared with fusion surgery, surgical decompression alone may be a suitable and less invasive option for the treatment of non-inflammatory retro-odontoid pseudotumor with AAS, especially in elderly patients.

Introduction

Non-neoplastic mass lesions adjacent to the odontoid process of the axis are known to be associated with rheumatoid arthritis [1,2] and hemodialysis [3,4]. On the other hand, retro-odontoid pseudotumors in patients without those conditions are often associated with non-inflammatory disorders such as atlantoaxial instability [5–10], which may result in cervical myelopathy requiring surgical intervention. Although direct excision and decompression of the pseudotumor by a transoral approach was proposed in the early days of retro-odontoid pseudotumor treatment, the risk of cerebrospinal fluid leakage, postoperative infection, and other complications was significant [11,12]. Surgeons have shifted to a minimally invasive approach of indirect decompression by upper cervical fusion and laminectomy [13].

Although posterior fusion and decompression is reportedly effective for retro-odontoid pseudotumor [6], good results with decompression surgery alone have been described as well [9,14]. Consensus on the optimal treatment for retro-odontoid pseudotumor is lacking, largely due to the limited number of reports comparing the procedures.

With the aging of the Japanese population, we have encountered more cases in which the invasiveness of fusion surgery poses a prohibitive risk. The present study compared the outcomes of posterior fixation with those of posterior decompression alone for relieving spinal cord pressure due to retro-odontoid pseudotumor with atlanto-axial subluxation (AAS). We aimed to determine whether decompression-only treatment could be an alternative to fusion surgery as a treatment for non-inflammatory retro-odontoid pseudotumor with AAS.

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Table 1
Characteristics of the study population.

	Fusion group	Non-fusion group	*p*-value*
	(*n* = 32)	(*n* = 9)	
Mean age (y)	71.8 ± 10.9	77.2 ± 12.5	0.26
Sex (male: female)	23: 9	4: 5	0.23
Observational period (months)	29.2 ± 16.5	22.2 ± 11.6	0.17
Comorbidities			
Hypertension [patients (%)]	14 (43.8)	3 (33.3)	0.71
Diabetes mellitus [patients (%)]	4 (12.5)	1 (11.1)	1
Cardiovascular disease [patients (%)]	3 (9.4)	0 (0)	1
Cerebrovascular disease [patients (%)]	5 (15.6)	1 (11.1)	1
Pulmonary disease [patients (%)]	1 (3.1)	1 (11.1)	0.39
Cancer [patients (%)]	3 (9.4)	1 (11.1)	1
Charlson Comorbidity Index (points)	0.50 ± 0.87	0.55 ± 0.72	0.84
Preoperative values			
JOA score	9.0 ± 3.2	8.2 ± 3.5	0.55
ADI (mm)	6.4 ± 2.5	5.6 ± 2.4	0.37
Change in ADI (mm)	3.7 ± 2.4	3.2 ± 2.7	0.61
C2–7 SVA (mm)	17.3 ± 17.0	22.3 ± 10.1	0.28
T1S (degrees)	23.0 ± 10.2	21.6 ± 7.5	0.65
CL (degrees)	12.8 ± 13.8	6.3 ± 8.0	0.09
T1S minus CL (degrees)	9.9 ± 12.6	15.2 ± 6.9	0.11
Perioperative values			
Surgical time (min)	230 ± 78	132 ± 65	0.002
Blood loss volume (mL)	217 ± 137	38 ± 64	< 0.001

All data are expressed as the mean ± standard deviation.
JOA: Japanese Orthopaedic Association, ADI: atlantodental interval, SVA: sagittal vertical axis, T1S: T1 slope, CL: cervical lordosis.
The change in ADI was defined as the difference in ADI at the flexion and extension positions.
*Determined by Welch's *t*-test or Fisher's exact test.

Materials and methods

Study population

This retrospective study was approved by our hospital's Investigational Review Board and included 41 patients (27 male and 14 female; mean age: 73.0 ± 11.4 years) who underwent posterior cervical fixation with or without additional decompression or decompression alone for the treatment of non-inflammatory retro-odontoid pseudotumor and were followed for more than 1 year between September 2009 and August 2019.

We diagnosed retro-odontoid pseudotumor and AAS by flexion lateral radiographs and MRI. The patients with cervical myelopathy due to retro-odontoid pseudotumor and atlantodental interval (ADI) of more than 3 mm in flexion lateral radiographs were included. Treatment was determined by consensus among several board-certified spine surgeons. Older patients and patients with more comorbidities tended to undergo decompression only. Rheumatoid arthritis patients, cerebral paralysis patients, patients without AAS, and patients with prior cervical spine surgery were excluded from this study.

We evaluated the result of treatments for retro-odontoid pseudotumor divided into patients who underwent posterior fusion surgery (fusion group) and those who received decompression surgery only (non-fusion group). The patient group characteristics are summarized in Table 1. Sex distribution and age were comparable between the fusion and non-fusion groups. There were no significant differences in comorbidities between the groups (Table 1). Mean scores for the Charlson Comorbidity Index [15] of classifying prognostic comorbidity in the fusion and non-fusion groups were comparable at 0.50 ± 0.87 points and 0.55 ± 0.72, respectively ($p = 0.84$) (Table 1). Preoperative ADI determined using preoperative flexion radiographs was 6.4 ± 2.5 mm in the fusion group and 5.6 ± 2.4 mm in the non-fusion group, which were similar ($p = 0.37$).

The change in ADI was defined as the difference in ADI at flexion and extension positions. ADI change was comparable between the fusion group (3.7 ± 2.4 mm) and the non-fusion group (3.2 ± 2.7 mm) ($p = 0.61$).

We also evaluated cervical alignment including parameters that included C2-7 sagittal vertical axis (SVA), T1 slope (T1S), cervical lordosis (CL), and T1S minus CL in lateral radiographs. Preoperative cervical alignment parameters and Japanese Orthopaedic Association (JOA) scores showed no remarkable differences.

Evaluation

The JOA scoring system for cervical myelopathy (full score: 17 points) was employed to evaluate clinical results preoperatively and at final follow-up. The JOA score evaluates the severity of cervical myelopathy and consists of 7 categories: motor function of the fingers, shoulder and elbow, and lower extremities, sensory function of the upper extremities, trunk, and lower extremities, and bladder function [16].

The JOA score recovery rate was calculated using the method described by Hirabayashi et al. [9] using the following equation: recovery rate (%) = (postoperative score – preoperative score) × 100 / (full score – preoperative score). The occurrence of perioperative and postoperative complications, operating time, and blood loss volume were recorded as well.

Data were analyzed by Welch's *t*-test for continuous data and Fisher's exact test for categorical data using EZR software (Saitama Medical Center, Jichi Medical University, Saitama, Japan), a graphical user interface for R (The Foundation for Statistical Computing, Vienna, Austria). The level of statistical significance was set at $p < 0.05$.

Results

Patient population

Thirty-two patients (23 male and 9 female; mean age: 71.8 ± 10.9 years) underwent posterior fusion surgery and 9 patients (4 male and 5 female; mean age: 77.2 ± 12.5 years) received decompression surgery

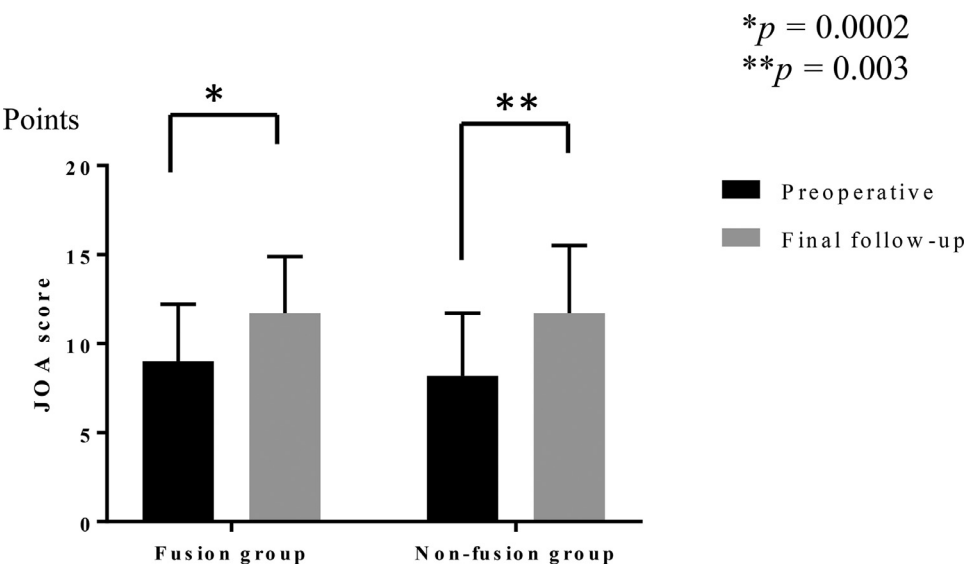

Fig. 1. Pre- and postoperative Japanese Orthopaedic Association (JOA) scores. In the fusion group, the mean preoperative JOA score improved significantly from 9.0 points to 11.7 points ($p = 0.0002$). Similarly in the non-fusion group, the mean preoperative JOA score improved significantly from 8.2 points to 11.7 points ($p = 0.003$).

only. In the fusion group, O-C fusion was completed in 12 patients and C1-2 fusion was performed in 20 patients. Eleven patients also underwent C1 laminectomy in the fusion group as a decompression procedure for cervical myelopathy caused by retro-odontoid pseudotumor with AAS.

Surgical variables

The average operating time in the fusion group (230 ± 78 min) was significantly greater than in the non-fusion group (132 ± 65 min) ($p = 0.002$). Similar findings were observed for mean blood loss (217 ± 137 mL vs. 38 ± 64 mL, respectively, $p < 0.001$) (Table 1).

The ADI in a neutral position at the final follow-up was 3.8 ± 1.8 mm in the fusion group and 3.7 ± 2.6 mm in the non-fusion group, which were statistically comparable ($p = 0.9$). Regarding complications, C5 palsy was observed in 1 patient in the fusion group who had received C3-C6 open-door laminoplasty. We did not perform laminoplasty of the middle or lower cervical spine in the other patients. Four patients in the fusion group experienced screw loosening, and 1 patient in the fusion group exhibited surgical site infection. None of the patients underwent reoperation.

Follow-up

The mean follow-up period of the cohort ranged from 12 to 62 months (mean: 27.3 ± 15.8 months). The mean follow-up in the fusion and non-fusion groups was similar at 29.2 ± 16.5 months (range: 12 to 62 months) and 22.2 ± 11.6 months (range: 12 to 48 months), respectively ($p = 0.17$).

Overall JOA scores in the cohort were significantly improved from 8.8 ± 3.2 points before surgery to 11.7 ± 3.2 points at the final follow-up ($p < 0.001$). In the fusion group, the respective mean preoperative and final follow-up JOA scores were 9.0 ± 3.2 points (range: 3 to 15.5 points) and 11.7 ± 3.2 points (range: 5 to 17 points), which indicated significant improvement ($p = 0.0002$) (Fig. 1).

Similarly in the non-fusion group, respective mean preoperative and final follow-up JOA scores showed significant gains from 8.2 ± 3.5 points (range: 1.5 to 13 points) to 11.7 ± 3.8 points (range: 4 to 17 points) ($p = 0.003$). The mean recovery rate of JOA score at the final follow-up was 22.6 ± 72.5% (range: −122 to 94.4%) in the fusion group and 43.4 ± 31.9% (range: 6.7 to 100%) in the non-fusion group, which were statistically comparable ($p = 0.23$) (Table 2).

Visual analog scale (VAS) scores for neck pain at the final follow-up were similar in the fusion group (49.0 ± 29.7 points [range: 0 to 100 points]) and the non-fusion group (63.3 ± 25.1 points [range: 30 to 100 points]) ($p = 0.17$) (Table 2).

Table 2
Comparison of JOA scores between the fusion and non-fusion groups.

	Fusion group	Non-fusion group	*p*-value*
	($n = 32$)	($n = 9$)	
Preoperative JOA score	9.0 ± 3.2	8.2 ± 3.5	0.55
Final follow-up JOA score	11.7 ± 3.2	11.7 ± 3.8	0.98
Recovery rate (%)	22.6 ± 72.5	43.4 ± 31.9	0.23
Final follow-up neck pain VAS	49.0 ± 29.7	63.3 ± 25.1	0.17

All data are expressed as the mean ± standard deviation.
JOA: Japanese Orthopaedic Association, VAS: visual analog scale.
*Determined by Welch's *t*-test.

Table 3
Correlations between cervical sagittal spinal alignment parameters and final follow-up JOA score recovery rate.

Parameter	Rho	*p*-value
C2–7 SVA	0.12	0.452
T1S	0.27	0.097
CL	0.14	0.396
T1S minus CL	0.13	0.431

JOA: Japanese Orthopaedic Association, SVA: sagittal vertical axis, T1S: T1 slope, CL: cervical lordosis.

The postoperative change in ADI was 0 mm in all patients in the fusion group. On the other hand, the mean postoperative ADI change was 2.4 ± 1.8 mm in the non-fusion group. In this group, the difference in ADI change between pre- and postoperatively was −0.7 ± 1.7 mm, with almost no increase in instability. Among the 9 patients in the non-fusion group, there was a slight increase in instability in 2 cases (22.2%; maximum increase: 1.2 mm).

We observed no significant correlations between any cervical sagittal spinal alignment parameter and JOA score recovery rate in the cohort (Table 3).

Case presentations

Case 1: A 66-year-old male was diagnosed as having cervical myelopathy with retro-odontoid pseudotumor and AAS. His preoperative JOA score was 13.5 points. Radiographs demonstrated cervical spondylosis and AAS (Fig. 2a-d). Preoperative ADI in the flexion position

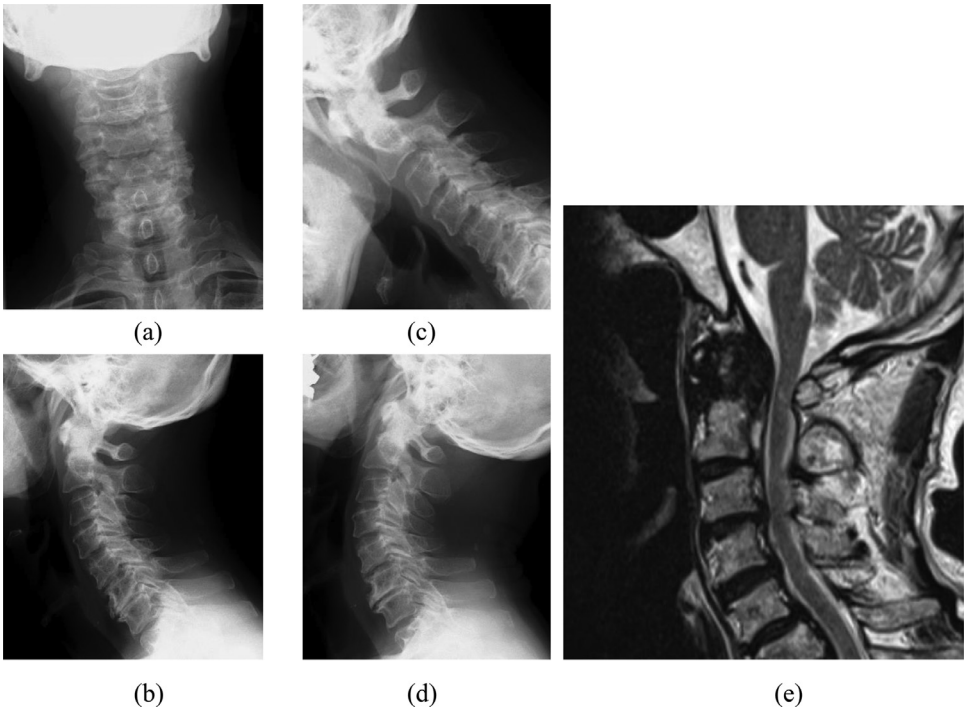

Fig. 2. Case 1: A 66-year-old male. a–d) Preoperative radiographs demonstrated cervical spondylosis and atlanto-axial subluxation. Preoperative atlantodental interval (ADI) in flexion was 5.2 mm and change in ADI was 3.7 mm. e) MRI revealed spinal cord compression by retro-odontoid pseudotumor.

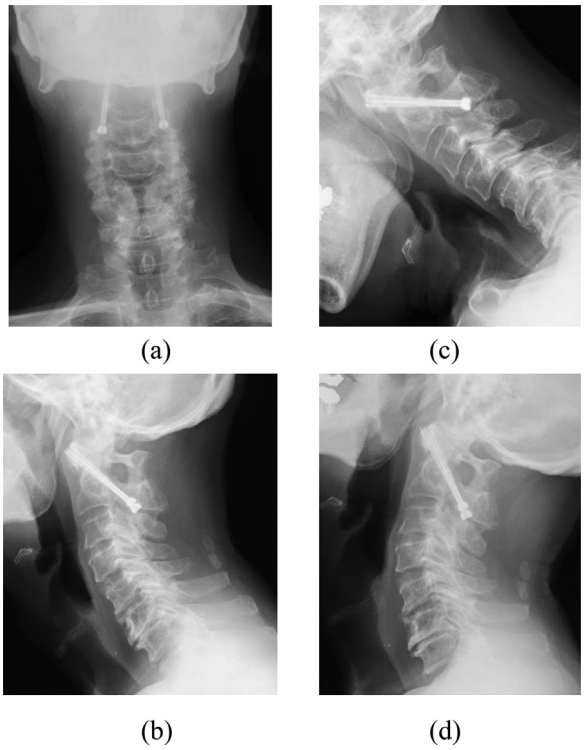

Fig. 3. Postoperative radiographs in Case 1. Atlantoaxial fusion was performed. Postoperative radiographs demonstrated no instability at C1–2.

was 5.2 mm and change in ADI was 3.7 mm (Fig. 2c, 2d). MRI revealed spinal cord compression by retro-odontoid pseudotumor (Fig. 2e). We performed atlantoaxial fusion with a Magerl and Brooks procedure. His postoperative JOA score was improved at 16.5 points at 5 years after surgery, with no apparent instability at C1-2 (Fig. 3).

Case 2: A 72-year-old female was diagnosed as having cervical myelopathy with retro-odontoid pseudotumor and AAS. Her preoperative JOA score was 7.5 points. Radiographs disclosed cervical spondylosis and AAS (Fig. 4a-–). Preoperative ADI during flexion was 4.0 mm and change in ADI was 1.6 mm (Fig. 4c, 4d). MRI revealed spinal cord compression by retro-odontoid pseudotumor (Fig. 4e). At 1 year after C1 laminectomy, her postoperative JOA score was improved at 15.5 points. Postoperative ADI in flexion was 4.0 mm and change in ADI was 1.6 mm (Fig. 5).

Discussion

In the present study, 41 patients with cervical myelopathy who underwent surgery for retro-odontoid pseudotumor with AAS were reviewed for comparisons of outcomes with posterior cervical fixation or decompression only. Both the fusion and non-fusion groups showed significant improvements, with comparable JOA score recovery rates and no major complications. Thus, the treatment options appeared similarly effective and safe in this investigation. However, retro-odontoid pseudotumors are more common in the elderly, who, given their many comorbidities, are already at a disadvantage in terms of recovery. If the non-fusion approach achieves the same benefits as fusion, decompression alone may be preferable due to its reduced invasiveness [17–20].

Retro-odontoid pseudotumors often cause progressive cervical myelopathy. Owing to the high risk of complications, pseudotumor treatment has shifted from tumor resection by a transoral approach to posterior fixation and decompression [11–13]. While some reports recommend fusion surgery due to the high frequency of AAS, other studies have described good results with surgical decompression alone [6,9,14]. Chikuda et al. [6] reported that approximately two-thirds of the retro-odontoid pseudotumor cases in the literature showed signs of atlantoaxial instability and were recommended a fusion procedure. On the other hand, Suetsuna et al. revealed that post-odontoid pseudotumors could regress spontaneously after C1 laminectomy [9]. Kakutani et al. described that C1 laminectomy in 7 patients with post-odontoid pseudotumor without AAS resulted in neurological improvement in all patients [14].

Currently, the literature on surgical outcomes in pseudo-tumor management is limited; the majority involve case series with a maximum of 30 patients and no clear evidence on which surgical approach is more

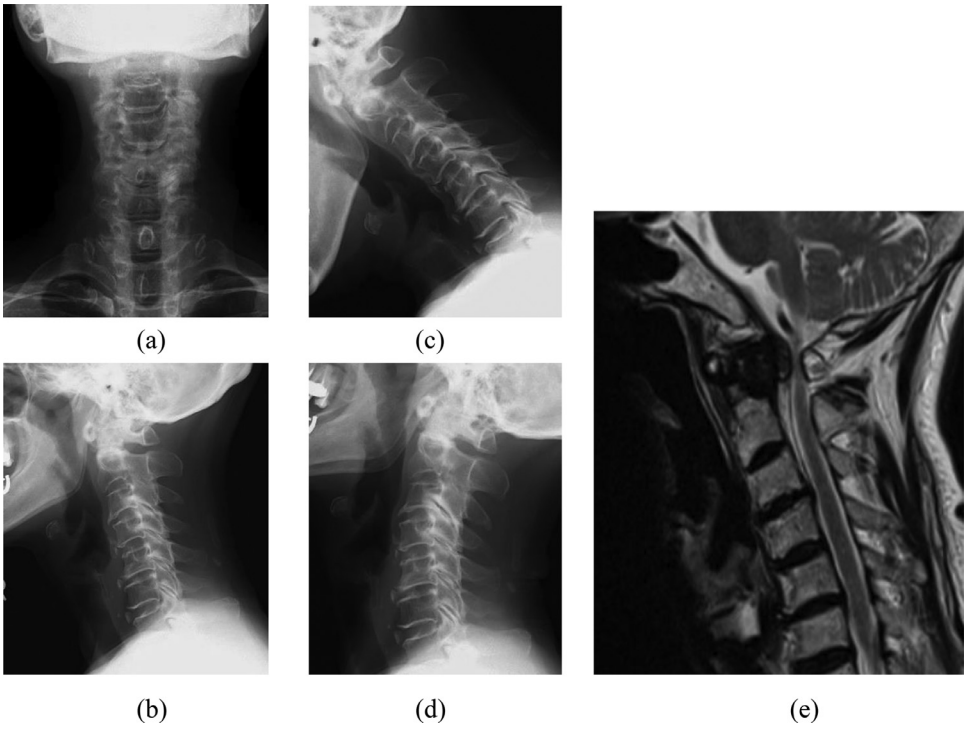

Fig. 4. Case 2: A 72-year-old female. a–d) Preoperative radiographs demonstrated cervical spondylosis and atlanto-axial subluxation. Preoperative atlantodental interval (ADI) in flexion was 4.0 mm and change in ADI was 1.6 mm. e) MRI revealed spinal cord compression by retro-odontoid pseudotumor.

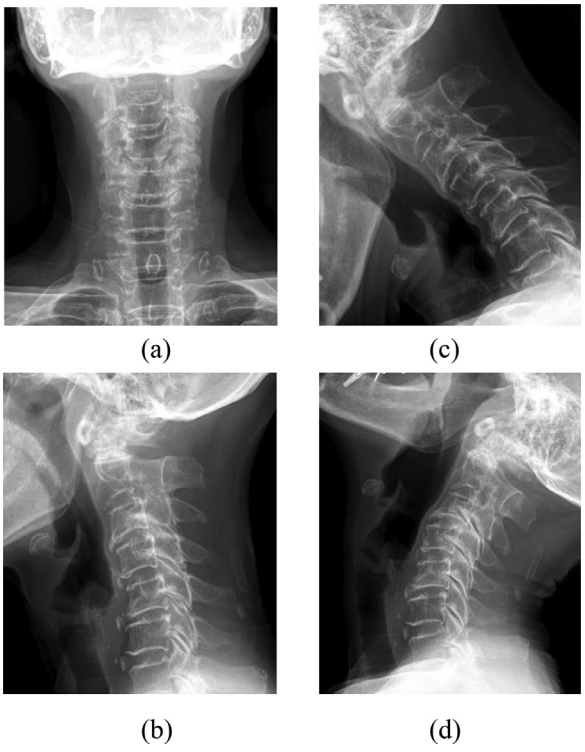

Fig. 5. Postoperative radiographs in Case 2. C1 laminectomy was performed. Postoperative atlantodental interval (ADI) in flexion was 4.0 mm and change in ADI was 1.6 mm.

appropriate, making the present cohort one of the largest to date [17–19,21–24]. Moreover, since patients with rheumatoid arthritis display most cases of retro-odontoid pseudotumor, there is a need for more data on non-inflammatory pseudotumors [21,22].

Elderly patients have a higher risk of surgical complications due to their generally greater prevalence of pre-existing medical conditions, such as cardiovascular complications. In recent years, it has become difficult to perform invasive fixation procedures effectively in the increasingly elderly population. Takeshima et al. revealed that elderly patients had a lower JOA score recovery rate in a meta-analysis of surgical outcomes in cervical spondylotic myelopathy patients [24]. In our study, both operative time and blood loss were significantly greater in the fusion group than in the non-fusion group. The patients in the non-fusion group tended to be older and therefore at a disadvantage in terms of symptom improvement. However, the JOA score recovery rate was similar between the groups. We consider that decompression alone for retro-odontoid pseudotumor with AAS is a reasonable procedure option, especially in the elderly.

Lastly, several studies have described a relationship between cervical spine alignment and cervical spine surgery outcomes [25–28]. Hyun et al. reported that a greater T1S minus CL mismatch was associated with worse neck disability index scores after posterior cervical fusion surgery [25,26], while Jeon et al. found that postoperative T1S minus CL and changes in T1S and T1S minus CL were significantly correlated with changes in neck disability index scores [27]. Chen et al. reported significant associations for C2-7 SVA and T1S minus CL with postoperative neck pain in cervical spinous process-splitting laminoplasty [28]. In our study, we observed no remarkable correlations between any cervical sagittal spinal alignment parameter and JOA score recovery rate. Moreover, VAS scores for neck pain were not significantly associated with C2-7 SVA (rho = 0.0623, $p = 0.702$) or T1S minus CL (rho = 0.122, $p = 0.447$).

This study had several limitations. Specifically, the follow-up period was relatively short and the number of cases was small. There remains a risk of instability and symptom worsening in the non-fusion group after time; however, in the 12 to 48 months of follow-up, all patients in the non-fusion group maintained improvement as compared with preoperative JOA scores. Considering the relative rareness of retro-odontoid pseudotumor and the small number of cases reported to date, it appears safe to assume that the number of cases in this study was sufficient for preliminary conclusions. The absence of a power analysis is also a limita-

tion, as is the lack of preoperative VAS data for neck pain. Lastly, there were no significant differences in demographic data or comorbidities between the groups, although treatment was determined by consensus among several board-certified spine surgeons.

In conclusion, the results of this comparative study indicated that surgical decompression without fusion could be a satisfactory and less invasive procedure for the treatment retro-odontoid pseudotumor with AAS in elderly patients with higher surgical risk.

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Declarations of Competing Interests

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.xnsj.2021.100064.

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