



Post-operative regression of retro-odontoid pseudotumors treated with and without fusion

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Abstract

Purpose Retro-odontoid pseudotumor is common in elderly people and is a cause of cervical myelopathy. The goal of the study was to investigate surgical procedures, outcomes, and post-operative spontaneous regression of posterior cervical retro-odontoid pseudotumors.

Methods The subjects were 29 patients who underwent surgery for myelopathy due to a retro-odontoid pseudotumor around the craniocervical region at 9 facilities and were followed-up for an average of 54 months (range 12–96 months). Data were collected in a multicenter review of a retrospective database. Comparisons were performed between cases treated with and without fusion.

Results The JOA recovery rate at final follow-up did not differ significantly between the fusion ($n = 17$, including all 15 patients with atlantoaxial subluxation) and non-fusion ($n = 12$) groups. However, pseudotumor regression was significantly more frequent in the fusion group (100% vs. 42%, $p < 0.01$). In all patients, regression cases had significantly higher rates of contrast enhancement of the pseudotumor on pre-operative T1 gadolinium-enhanced MRI (68% vs. 14%, $p = 0.013$) and of JOA recovery (50% vs. 30%, $p < 0.01$).

Conclusions Regression of pseudotumor occurred in all cases treated with fusion surgery. There was a significant difference in pseudotumor regression with or without fusion, and regression was significantly related to gadolinium enhancement on MRI. Therefore, it is preferable to use fusion surgery for a retro-odontoid pseudotumor that shows contrast enhancement, even if there is no apparent instability pre-operatively.

Graphical abstract These slides can be retrieved under Electronic Supplementary Material.

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Key points

1. Outcomes for posterior cervical retro-odontoid pseudotumors treated surgically with and without fusion were investigated in a multicenter study.
2. Outcomes (JOA scores) did not differ significantly between fusion and non-fusion surgeries, but pseudotumor regression was significantly more frequent after fusion.
3. Pseudotumor regression was significantly related to JOA recovery rate at final follow up.
4. Pseudotumor regression was significantly related to gadolinium enhancement on MRI.
5. A retro-odontoid pseudotumor with contrast enhancement should preferably be treated with fusion surgery to promote pseudotumor regression, even if there is no apparent instability before surgery.

[Citation]
Chikuda H, Seichi A, Takeshita K et al (2009) Radiographic analysis of the cervical spine in patients with retro-odontoid pseudotumors. Spine 34:E110–E114

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Parameter	Fusion (n=17)	Non-fusion (n=12)	p-value
JOA recovery rate (%)	50	30	<0.01
Pseudotumor regression (%)	100	42	<0.01
Contrast enhancement on MRI (%)	68	14	0.013

[Citation]
Yamaguchi I, Shibuya S, Arima N et al (2006) Remarkable reduction or disappearance of retroodontoid pseudotumors after occipitocervical fusion. Report of three cases. J Neurosurg Spine 2006;5:156–160

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Take Home Messages

1. Regression of pseudotumor occurred in all cases treated with fusion surgery.
2. There was a significant difference in pseudotumor regression with or without fusion, and regression was related to gadolinium enhancement on MRI.
3. It is preferable to use fusion surgery for a retro-odontoid pseudotumor that shows contrast enhancement.

[Citation]
Kakutani K, Doita M, Yoshikawa M et al (2013) C1 laminectomy for retro-odontoid pseudotumor without atlantoaxial subluxation: review of seven consecutive cases. Eur Spine J 22:1119–1126

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Keywords Retro-odontoid pseudotumor · Atlantoaxial instability · Occipitocervical fusion · Posterior decompression · Spinal fusion · Gadolinium-enhanced MRI

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Introduction

Retro-odontoid pseudotumor occurs in many elderly people and causes cervical myelopathy as a space-occupying lesion [1, 2]. The pseudotumor is mainly associated with atlantoaxial subluxation (AAS) and mechanical stress [1, 3, 4]. Pathologically, retro-odontoid pseudotumor is composed of fibrous granulation or fibrocartilaginous tissue [1, 5–7]. Rheumatoid arthritis (RA), long-term hemodialysis, migrated disc herniation, and mucinous degeneration of the transverse ligament of the atlas are possible causes [8–11].

Development of chronic atlantoaxial instability and resulting mechanical stress are treated surgically using posterior fusion with occipitocervical or atlantoaxial fusion to prevent mechanical stress on the atlantoaxial joint. This procedure is an appropriate surgical strategy for retro-odontoid pseudotumor with atlantoaxial subluxation [1, 4, 7, 12–16], and neurological and symptomatic improvements are likely after posterior fusion [4, 13]. Spontaneous regression of pseudotumors following posterior fusion has also been reported [1, 7]. C1 laminectomy also has efficacy and may lead to spontaneous regression of retro-odontoid pseudotumors [2, 17].

There have been no comparative studies of fusion and non-fusion procedures focused on regression of retro-odontoid pseudotumors. Therefore, the purpose of this study is to investigate surgical procedures and outcomes for posterior cervical retro-odontoid pseudotumors, including spontaneous regression, following surgery at multiple centers.

Materials and methods

The subjects were 29 patients (23 males and 6 females) who underwent surgery for myelopathy due to a retro-odontoid pseudotumor in the craniocervical region at 9 facilities associated with our group. Data were examined retrospectively. Age ranged from 54 to 85 years, with a mean of 74.4 years. The average follow-up period was 54 months (range 12–96 months). The study was approved by the Ethics Committee of our hospital (IRB No. 354-3).

Diagnosis of a retro-odontoid pseudotumor was made using magnetic resonance imaging (MRI), including gadolinium (Gd) enhancement of a mass lesion posterior to the odontoid process with substantial cord compression. Lesions were classified into three types using the classification of Yonezawa et al., which is based on the signal intensity on T2-weighted images (T2WI): high intensity as a pannus, low intensity as a pseudotumor, and mixed low

and high intensity as mixed type [16]. The maximal thickness of the retro-odontoid soft tissue was measured on pre-operative and post-operative T2WI in the sagittal view. The reduction rate of each pseudotumor was calculated as: [(maximum pre-operative thickness – maximum post-operative thickness)/maximum pre-operative thickness] × 100. A reduction rate > 50% was defined as regression.

Radiographs of the cervical spine were obtained in all patients in the neutral, flexion, and extension positions. The atlantodental interval (ADI) was measured as Δ ADI in pre-operative lateral flexion–extension radiographs. AAS was defined as Δ ADI > 4 mm, using the criteria described by White et al. [18]. Range of motion (ROM) was measured on pre-operative flexion–extension radiographs. The lines used for measurements were as follows: McGregor line, the line passing through the centers of the anterior and posterior arches of the atlas, and the line parallel to the endplate of vertebrae. Radiographic measurements were performed independently by two spine surgeons. Sagittal computed tomography (CT) reconstructions were evaluated for the presence of ossification of the anterior longitudinal ligament (OALL), which was defined as a bony mass anterior or posterior to vertebrae bridging over two or more intervertebral disc spaces.

C1–2 fusion was performed in all cases of AAS. Fusion surgery was performed in cases with OALL, occipito annular fusion, and retro-odontoid cyst, in addition to cases with atlantoaxial subluxation (Fig. 1). The fusion ranges were chosen based on the presence of ligament ossification, regional kyphosis, and bone fusion. If compression was severe in the posterior spinal cord due to a C1 pseudotumor, we planned to add C1 laminectomy. The severity of myelopathy was assessed using the Japanese Orthopaedic Association (JOA) scoring system [19]. Surgical outcomes were evaluated using the Hirabayashi recovery rate [(post-operative JOA score – pre-operative JOA score)/pre-operative JOA score]. RA was diagnosed according to 1987 American Rheumatism Association criteria [20]. For assessment of regression of pseudotumor, patients were divided into those treated with and without fusion; and pre- and post-operative parameters were investigated in these groups. Differences between groups were analyzed by Mann–Whitney *U* test or Student's *t* test. All statistical analyses were conducted using SPSS ver. 22 for Windows (IBM, Chicago, IL). *p* < 0.05 were considered to be significant in all analyses.

Results

Demographic data are shown in Table 1. The 29 patients included 5 with RA; 3 on hemodialysis; 12 with cervical spondylotic myelopathy; and 15 with AAS, including 4 with RA. On pre-operative MRI, 15 patients, including 3

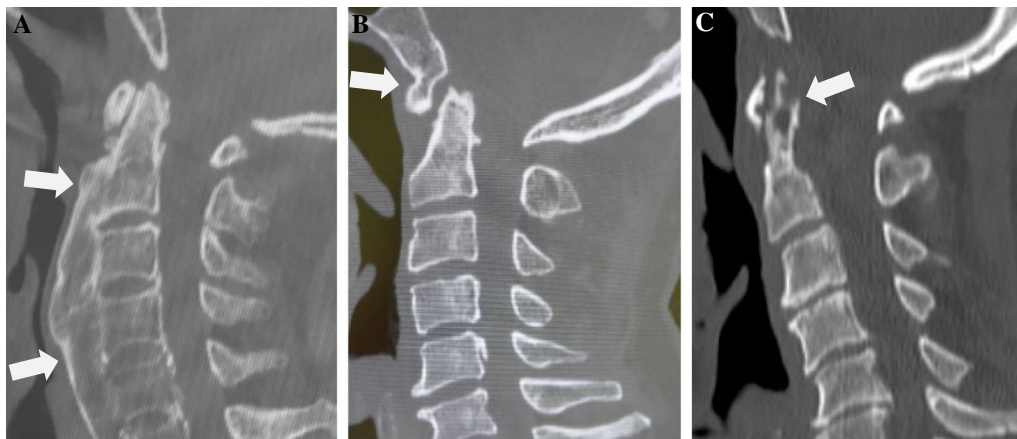


Fig. 1 Cases with OALL (a), occipito annular fusion (b), and retro-odontoid cyst (c). Arrows indicate the lesion

Table 1 Demographic and clinical characteristics in patients treated with and without fusion ($n=29$)

Item	Fusion ($n=17$)	No fusion ($n=12$)	<i>p</i>
Demographics			
Age (years)	72.5 ± 9.2	77.5 ± 4.8	n.s.
Female (n)	3 (18%)	3 (25%)	n.s.
Follow-up period (months)	54.3 ± 30.6	49.8 ± 35.2	n.s.
Etiology			
Rheumatoid arthritis (n)	4 (24%)	1 (8%)	n.s.
Hemodialysis (n)	2 (12%)	1 (8%)	n.s.
OALL (n)	5 (29%)	1 (8%)	n.s.
Atlantoaxial subluxation (n)	15 (88%)	0 (0%)	<0.01
MRI (T1-weighted/T2-weighted)			
Low/low	10 (58%)	5 (42%)	n.s.
Low/high	4 (24%)	3 (25%)	n.s.
Low/low + high	3 (18%)	4 (33%)	n.s.
MRI (gadolinium administration)			
Enhance	11 (65%)	5 (42%)	n.s.
Surgical procedures			
Resection of C1 posterior arch	5 (29%)	12 (100%)	<0.01

on hemodialysis, had low signal intensities on T1WI and T2WI; 7, including 3 with RA, had low signal intensity on T1WI and high signal intensity on T2WI; and 7 had low signal intensity on T1WI, and low and high signal intensity on T2WI.

Posterior fusion was performed in 17 patients, at O–C2 ($n=3$), O–C4 ($n=2$), O–C5 ($n=2$), and C1–2 ($n=10$, including 2 patients with a C1–2 transarticular screw and 8 with a C1 lateral mass and C2 pedicle screw). The 17 fusion cases included 12 treated without C1 posterior arch resection and 5 who underwent C1 posterior arch resection. In the non-fusion group ($n=12$), C1 posterior arch resection was performed alone in eight cases and in combination with middle or lower cervical laminoplasty in 4, while transoral decompression was not performed (Fig. 2).

Post-operative MRI at final follow-up showed regression of pseudotumor in 22 patients, included 17 treated with fusion surgeries and 5 with non-fusion surgeries. All fusion cases showed pseudotumor regression. The mean pre- and post-operative JOA scores were 7.7 and 11.7 in the fusion group ($n=17$), and 8.3 and 12.2 in the non-fusion group ($n=12$). All patients showed neurological recovery, and the respective mean recovery rates in the two groups were 51 and 48%, with no significant difference between the groups. However, the pseudotumor regression rate was significantly higher in the fusion group (100% vs. 42%, $p<0.01$) (Table 2).

The regression group had significantly higher rates of fusion surgery among all surgeries (77% vs. 0%, $p<0.01$), JOA recovery rate (50% vs. 30%, $p<0.01$), and pre-operative

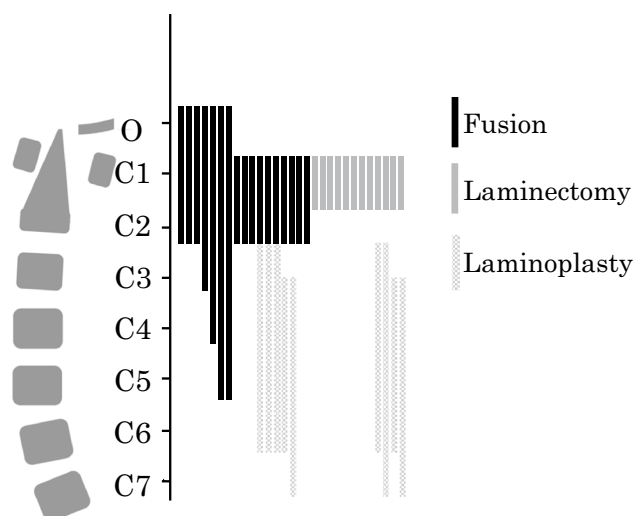


Fig. 2 Range and surgical procedure in the 29 cases

Table 2 Comparison of pre-operative and post-operative status in patients treated with and without fusion ($n = 29$)

Item	Fusion ($n = 17$)	No fusion ($n = 12$)	p
Pre-operative atlantodental interval			
Flexion (mm)	7.2 ± 2.8	3.2 ± 0.7	<0.01
Extension (mm)	2.6 ± 0.6	2.4 ± 0.6	n.s.
Δ ADI (mm)	4.6 ± 1.9	0.7 ± 0.4	<0.01
Range of motion ($^{\circ}$)			
O–C2	18.6 ± 11	20.3 ± 9	n.s.
C2–C7	22.3 ± 13	18.7 ± 11	n.s.
JOA score			
Pre-op score	7.7 ± 2.9	8.3 ± 1.7	n.s.
Post-op score	11.7 ± 4.1	12.2 ± 2.7	n.s.
Recovery rate (%)	51 ± 32	48 ± 31	n.s.
Follow-up MRI			
Regression (n)	17 (100%)	5 (42%)	<0.01
Regression rate (%)	73 ± 8	48 ± 17	<0.01
Reoperation (n)	0	1	n.s.

Δ ADI indicates difference between flexion and extension

Gd contrast enhancement of the pseudotumor on T1 MRI (68% vs. 14%, $p = 0.013$) (Table 3).

Illustrative cases

Case 1: O–C3 fusion

A 72-year-old man visited our hospital due to gait disturbance and neck pain. The JOA score was 6 at the first visit. MRI showed a high T2WI signal in the spinal cord at the C1 level with severe spinal canal stenosis due to a retro-odontoid mass lesion, and pseudotumor was confirmed on

Table 3 Comparison of cases with and without pseudotumor regression in follow-up MRI ($n = 29$)

Item	Regression (+) ($n = 22$)	Regression (–) ($n = 7$)	p
Etiology			
Rheumatoid arthritis (n)	4	1	n.s.
Hemodialysis (n)	2	1	n.s.
OALL (n)	5	1	n.s.
Atlantoaxial subluxation (n)	15	0	<0.01
MRI (T1-weighted/T2-weighted)			
Low/low	12 (55%)	3 (43%)	n.s.
Low/high	6 (27%)	1 (14%)	n.s.
Low/low + high	4 (18%)	3 (43%)	n.s.
MRI (gadolinium administration)			
Enhancement	15 (68%)	1 (14%)	0.013
Surgical procedures			
Fusion (n)	17 (77%)	0 (0%)	<0.01
JOA score			
Pre-op score	8.3 ± 1.8	8.7 ± 2.1	n.s.
Post-op score	12.2 ± 2.3	11.3 ± 2.9	n.s.
Recovery rate (%)	50 ± 27	30 ± 7	<0.01

Gd-enhanced MRI. CT showed a retro-odontoid cyst. Thus, we performed fusion surgery in the O–C3 range. Post-operative T2WI showed that the mass lesion had regressed at 1 year after posterior fusion surgery. Post-operative myelopathy improved markedly, and the JOA score had improved from 6 to 11 points (recovery rate: 83%) at 1 year after surgery (Fig. 3).

Case 2: C1 laminectomy (non-fusion)

A 67-year-old man who had undergone hemodialysis for 9 years visited our hospital due to muscle weakness in the upper extremities. The JOA score was 5 at the first visit. MRI showed a retro-odontoid mass lesion and subaxial degenerative changes. Pseudotumor was not confirmed on Gd-enhanced MRI. Lateral extension–flexion radiographs did not show AAS. C1 laminectomy with C2–6 laminoplasty was performed. Post-operatively, motor weakness recovered and the JOA score was 10 at 1 year after surgery. However, MRI at 1 year post-operatively did not show regression of the pseudotumor (Fig. 4).

Discussion

A pseudotumor may be caused by a benign fibrous mass formed as a retro-odontoid due to mechanical stress caused by chronic AAS [3]. Crockard et al. suggested that the pseudotumor is formed by repetition of partial rupture

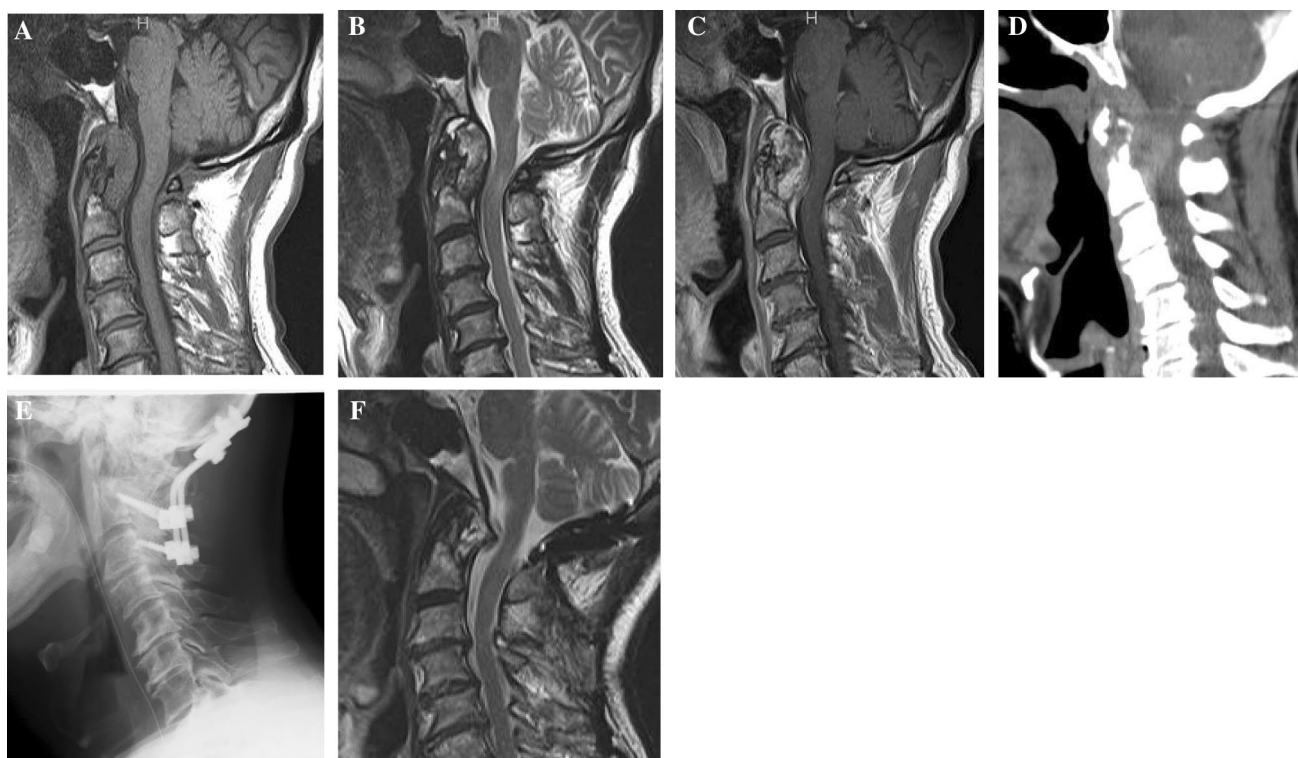


Fig. 3 Case 1: **a, b** retro-odontoid pseudotumor detected on sagittal T1- (**a**) and T2-weighted MRI (**b**). **c, d** Capsular enhancement shown by Gd-enhanced MRI (**c**). **d** Sagittal CT showing a retro-odontoid

cyst. **e** Post-operative lateral radiography after surgery with O–C3 fusion. **f** Post-operative image 1 year after surgery showing regression of the pseudotumor

and repair in the atlantocruciate ligament due to cervical spondylotropic changes, even without obvious instability in the atlantoaxial joint [6]. Shah et al. suggested that pseudotumor could be a manifestation of atlantoaxial instability [21], and Chikuda et al. found that the transverse ligament is damaged from excessive stress on the atlantoaxial joint and forms a mass in reaction [7]. On the other hand, Bydon et al. reported that rheumatoid patients develop a retrodental lesion (atlantoaxial rheumatoid pannus) with cervical instability [22], and Finn et al. described pseudotumor accompanied with atlantoaxial degenerative arthritis [23]. Therefore, the suggested pathogenesis is mechanical dysfunction and instability of the atlantoaxial joint that promotes development of hypertrophic soft tissue in the retro-odontoid area.

Transoral decompression is an ideal surgery for pseudotumors, because it allows for direct excision of a compressive mass [3, 6], but it is also associated with significant complications [24, 25], including bacterial infection, cerebrospinal fluid leakage, and post-operative instability of the atlantoaxial joint. A posterior approach is currently the primary choice, and there have been several reports of pseudotumor regression by posterior decompression and fusion [2, 7, 12, 15, 16, 22, 26, 27, 28, 29] (Table 4). Barbagallo et al. suggested that a posterior approach might be sufficient and that transoral surgery was not required [27]. We have

also used posterior fusion and obtained satisfactory clinical outcomes with adequate regression of the pseudotumor in all cases. We believe that it is preferable to perform atlantoaxial fusion, given that the cause of the pseudotumor is instability of the annular joint. In many AAS cases, there is subarachnoid space behind the C1 pseudotumor, which makes it unnecessary to use C1 laminectomy. Therefore, C1–2 fixation without C1 laminectomy is preferable. However, if compression of the posterior spinal cord is severe due to a C1 pseudotumor, C1 laminectomy can be added to fusion surgery.

For cases with no obvious instability of the atlantoaxial joint, C1 posterior arch resection alone can improve clinical symptoms [1, 2, 15, 30] and may lead to pseudotumor regression [2, 31]. Takemoto et al. found that C1 laminectomy alone did not cause severe AAI progression and no patients showed serious mass enlargement [29]. However, Arima et al. described posterior arch resection for cases with no atlantoaxial joint instability, with reoperation for O–C fusion in one case because of an increase in pseudotumor mass and deterioration of symptoms after surgery [26]. In our series, C1 laminectomy produced satisfactory clinical outcomes, similar to posterior fusion surgery, but, in the non-fusion group, there were seven cases without pseudotumor regression.

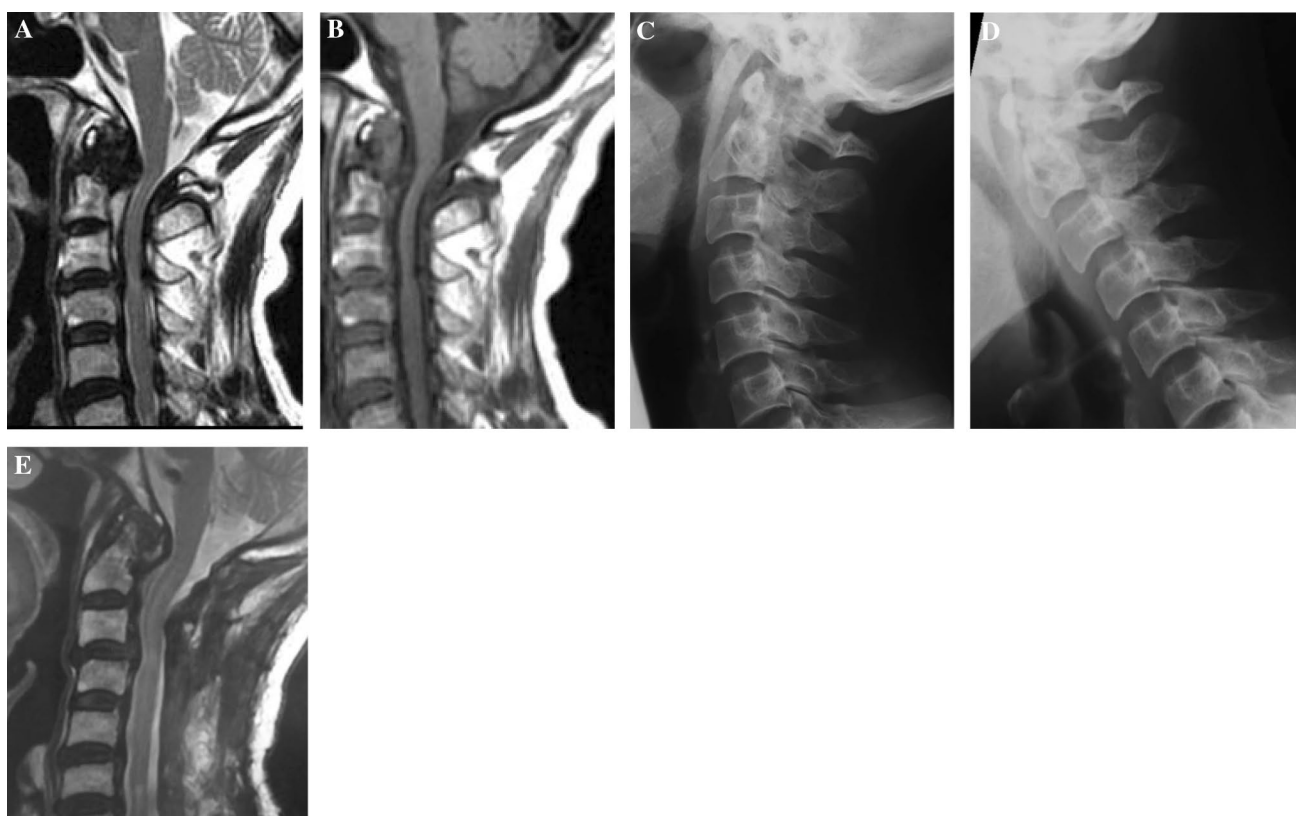


Fig. 4 Case 2: **a** retro-odontoid pseudotumor detected on T2-weighted MRI. **b** Enhancement was not present on Gd-enhanced MRI. **c, d** Lateral extension–flexion radiographs did not show AAS (Δ ADI=2.2 mm). **e** Image at 1 year after surgery, showing no regression of the retro-odontoid pseudotumor

Table 4 Review of case series with retro-odontoid pseudotumor treated by a posterior approach

Year	Author	No. of fusion/non-fusion	Regression rate of follow-up MRI (%) fusion/non-fusion	Follow-up (months)	Relationship of pre-operative MRI with post-operative pseudotumor regression
1997	Grob et al. [12]	22 fusions	100% (22/22)	NA	NA
2009	Chikuda et al. [7]	9 fusions/1 non-fusion	78% (7/9)/100% (1/1)	30	NA
2013	Arima et al. [26]	3 fusions/2 non-fusion	67% (2/3)/50% (1/2)	30	NA
2013	Yonezawa et al. [16]	11 fusions	100% (11/11)	NA	Pannus (high intensity on T2 and low intensity on T1) disappeared within 1 month of surgery. Low intensity on T2 disappeared a few months later
2013	Kakutani et al. [2]	7 non-fusion	57% (4/7)	52	80% of Gd enhancement of pseudotumor had complete regression
2013	Barbagallo et al. [27]	5 fusions	100% (5/5)	32	NA
2014	Sono et al. [28]	11 fusions	91% (10/11)	52	NA
2015	Bydon et al. [22]	30 fusions	100% (30/30)	24	NA
2015	Tominaga et al. [15]	4 fusions/1 non-fusion	100% (4/4)/0% (0/1)	35	Pseudotumor with mixed low and high intensity on T2 did not change or slow regression
2016	Takemoto et al. [29]	10 non-fusions	40% (4/10)	29	NA
2017	This study	17 fusions/12 non-fusions	100% (17/17)/42% (5/12)	54	Gd enhancement of the pseudotumor on pre-operative MRI was significantly higher in regression cases (68% vs. 14%, $p=0.013$)

NA not available, Gd gadolinium

In our series, the JOA recovery rate showed no significant difference between the fusion and non-fusion groups. This may be because the pre-operative JOA score was higher in the non-fusion group; that is, the pre-operative symptoms were not as severe in this group. In addition, the JOA score recovery rate was significantly less in no pseudotumor regression of non-fusion group. Taking into account that only 42% of non-fusion cases had pseudotumor regression, improvement of post-operative symptoms was inferior to that in no regression group, and pseudotumor regression was achieved in all cases with fusion surgery, we believe that we do not always recommend non-fusion surgery with low tumor regression rate, but seeking the indication for fusion surgery for achieving pseudotumor regression.

In our series, pseudotumor regression was significantly associated with Gd enhancement of the pseudotumor on MRI. These findings suggest that although retro-odontoid pseudotumor is mostly composed of fibrous granulation or fibrocartilaginous tissue, angiogenesis may have been related to regression of the pseudotumor, based on the Gd enhancement. Kakutani et al. previously reported that regression of pseudotumors after C1 laminectomy is related to contrast enhancement on MRI after Gd administration, with 80% (4/5 cases) with Gd enhancement of a pseudotumor having complete post-operative regression (Table 4). This enhancement is thought to reflect neovascularization around the pseudotumor. These are consistent with our results. Cases without AAS, especially those with Gd enhancement on MRI, are predisposed to spontaneous regression after C1 laminectomy [2], and our findings are consistent with this result. To the best of our knowledge, the current study is the largest to examine the relationship of pre-operative MRI findings with post-operative pseudotumor regression (Table 4).

Chikuda et al. suggested that the cause of pseudotumor is not only related to AAS, but is also strongly related to OALL and adjacent vertebral body union of the cervical vertebrae; thus, fusion surgery was recommended even in cases without AAS [7]. Wakatsuki et al. reported that assimilation of the atlas is likely to occur at a high rate in AAS [30]. In our series, pseudotumor regression occurred after fusion surgery in non-AAS cases of OALL, craniocervical fusion, and retro-odontoid cyst, consistent with the previous reports. Thus, fusion surgery is preferable in such cases.

Crowned Dens Syndrome (CDS) should be mentioned as a differential diagnosis. CDS was first described by Bouvet et al. in 1985 [32] as a clinical-radiologic entity consisting of neck pain and cervical spine calcium crystal deposition, with CT showing densities surrounding the top and sides of the odontoid process in a crown- or halo-like distribution. Neck pain is mainly due to calcifications surrounding the odontoid process, including the transverse ligament of the atlas and ligamentum flavum [33]. CDS

is an important consideration in differential diagnosis of neck pain and mimics retro-odontoid pseudotumor.

The limitations of the study include the small number of cases, the non-prospective design, the variety of surgical procedures, including cervical laminoplasty due to degenerative changes, and variations in the protocol in a multicenter study. However, MRI including Gd enhancement was performed post-operatively in all cases and full clinical records were available, which allowed pseudotumor regression to be evaluated. This is also the first comparative study of treatment with or without fusion for retro-odontoid pseudotumor focused on pseudotumor regression, and we believe that the results provide important information for surgical planning.

In conclusion, we have described cases of retro-odontoid pseudotumor in which marked regression of the pseudotumor occurred after surgical decompression and fusion without direct pseudotumor excision. All cases treated with fusion showed regression of the pseudotumor, but without a significant difference in JOA recovery rate in comparison with C1 laminectomy. These findings suggest that C1 laminectomy is suitable for patients without AAS and can improve clinical outcomes. However, some C1 laminectomy cases did not show a significant pseudotumor regression post-operatively. Furthermore, in non-AAS cases that underwent C1 laminectomy, pseudotumor regression was significantly associated with JOA score recovery and Gd enhancement of the pseudotumor on pre-operative MRI. Thus, even in cases with no obvious instability of the atlantoaxial joint, it is preferable to perform fusion surgery if the pseudotumor shows Gd enhancement on MRI.

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Compliance with ethical standards

Conflict of interest None of the authors have a conflict of interest.

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