# ORIGINAL ARTICLE

# C1 laminectomy for retro-odontoid pseudotumor without atlantoaxial subluxation: review of seven consecutive cases

Kenichiro Kakutani · Minoru Doita · Masaho Yoshikawa · Koji Okamoto · Koichiro Maeno · Takashi Yurube · Norihide Sha · Masahiro Kurosaka · Kotaro Nishida

Received: 27 October 2012/Revised: 7 January 2013/Accepted: 22 January 2013/Published online: 6 February 2013 © Springer-Verlag Berlin Heidelberg 2013

#### Abstract

*Purpose* A retro-odontoid pseudotumor is usually a reactive fibrocartilaginous mass associated with atlantoaxial subluxation (AAS). However, a retro-odontoid pseudotumor not associated with AAS, which undergoes spontaneous regression following C1 laminoplasty, has been reported. The purpose of this study was to report surgical outcomes of C1 laminectomy for retro-odontoid pseudotumor without AAS.

*Materials and methods* The cases of seven patients (mean age  $75.6 \pm 7.6$  years-old) with retro-odontoid pseudotumor without AAS were reviewed. The mean follow up time was  $52.3 \pm 25.5$  months. Each patient underwent a C1 laminectomy with an additional C3–6 expansion laminoplasty in three patients. The Japanese Orthopaedic Association score (JOA score) was used for neurological assessment. Pseudotumor size and additional AAS were analyzed using MRI and radiography.

*Results* All patients exhibited neurological improvement following surgery, the JOA score improved from  $7.2 \pm 3.2$ to  $14.1 \pm 2.6$ . The mean O-C2 and C2–7 angle decreased from  $-3.2 \pm 2.1^{\circ}$  to  $-3.9 \pm 1.7^{\circ}$ , showing a slight kyphotic change. Postoperative AAS was not observed. All pseudotumors spontaneously resolved, and recurrence and regrowth were not observed. Five patients had MRIs after gadolinium administration; four patients who showed enhancement of the pseudotumor had almost complete reduction within 1 year following surgery.

K. Maeno  $\cdot$  T. Yurube  $\cdot$  N. Sha  $\cdot$  M. Kurosaka  $\cdot$  K. Nishida

Department of Orthopaedic Surgery, Kobe University Graduate School of Medicine, 7-5-2 Kusunokicho,

Graduate School of Medicine, 7-3-2 Kusunokicho,

Chuo-ku, Kobe 650-0017, Japan

*Discussion* Our study, assessing the outcome of C1 laminectomy for retro-odontoid pseudotumor, found neurological improvement in all cases. Since all pseudotumors were reduced and additional AAS was not observed, C1 laminectomy for retro-odontoid pseudotumor, in the absence of AAS, is recommended as a therapeutic strategy.

**Keywords** Retro-odontoid pseudotumor · Spontaneous regression · C1 laminectomy

## Introduction

The increased use of magnetic resonance imaging (MRI) for assessing lesions of the craniovertebral junction has allowed non-neoplastic mass lesions adjacent to the odontoid process of the axis to be increasingly diagnosed. These non-neoplastic mass lesions are known to be associated with rheumatoid arthritis [1, 2], and with hemodialysis [3, 4], frequently leading to cervical myelopathy. The masses in the posterior odontoid process, not associated with the underlying disease, have been reported to be reactive fibrocartilaginous masses induced by chronic subluxation of the craniovertebral junction [5, 6]. Preexisting atlantoaxial instability has been considered to cause repeated tears and subsequent hypertrophy of the transverse ligament, leading to retro-odontoid pseudotumor [7, 8]. However, retro-odontoid pseudotumors not associated with any apparent atlantoaxial (AAS) subluxation are occasionally encountered [9]. Ten cases of retro-odontoid pseudotumor without AAS were reported by Chikuda et al. They found that extensive ossification of the anterior longitudinal ligament (OALL) and ankylosis of the adjacent segments were risk factors for the formation of pseudotumors [10]. More specifically, the altered biomechanics of

K. Kakutani ( $\boxtimes$ ) · M. Doita · M. Yoshikawa · K. Okamoto ·

e-mail: kenkakutani@gmail.com; kakutani@med.kobe-u.ac.jp

the upper cervical spine due to ankylosis caused by OALL, ossification of the posterior longitudinal ligament (OPLL) and cervical kyphosis might load mechanical stress on the atlantoaxial joint, leading to development of a retro-odontoid pseudotumor without atlantoaxial subluxation.

Based on this proposed etiology of pseudotumors, posterior fusion with occipitocervical or atlantoaxial fusion to prevent mechanical stress to the atlantoaxial joint has been considered to be an appropriate surgical strategy for retro-odontoid pseudotumor with/without atlantoaxial subluxation. Neurological and symptomatic improvement can be expected following posterior fusion [8, 11, 12]. Furthermore, spontaneous regression of pseudotumors following posterior fusion has been reported [10, 13]. On the other hand, Suetsuna et al. [14] reported spontaneous regression of retro-odontoid pseudotumors following C1 laminoplasty. The etiology of retro-odontoid pseudotumor remains controversial and the appropriate surgical procedure to treat it has not yet been established.

In this study, the authors retrospectively examined the effects of C1 laminectomy on seven patients with retroodontoid pseudotumor without concurrent atlantoaxial subluxation. The purpose of the study was to investigate the clinical efficacy of C1 laminectomy for retro-odontoid pseudotumors and to determine radiographic changes, including spontaneous regression of pseudotumors and realignment of the cervical spine following surgery.

## Materials and methods

#### Patients and surgery

After approval by the institutional review board, we reviewed the clinical records of seven patients with cervical myelopathy due to retro-odontoid pseudotumor who had been surgically treated at our institutions. These patients were followed for more than 2 years [mean  $\pm$  standard deviation (SD)  $52.3 \pm 25.5$  months, range 27.7–99.0 months]. There were six men and one woman and the mean age at the time of surgical intervention was 75.6  $\pm$  7.6 yearsold (range 64-87 years-old). Patients with retro-odontoid reactive lesion associated with cervical trauma and systemic disorders, including rheumatoid arthritis and hemodialysis, were excluded from the study. The neurological findings, bilateral scapulohumeral reflex, hyperreflexia, positive pathological reflexes, motor weakness and sensory disturbance, revealed that all patients had cervical myelopathy due to retro-odontoid pseudotumors.

For this study, a retro-odontoid pseudotumor was defined as a mass lesion existing in the retro-odontoid space, which was hypointense on T1-weighted images and

from hypointense to hyperintense on T2 weighted images and which did not show any calcified component revealed by CT scan. MRI after gadolinium (Gd) administration was performed in only five of the patients because two patients who had mild renal failure were excluded. Because no patients had instability in the atlantoaxial joint or the cervical spine, all seven were treated by C1 laminectomy. Three patients who showed cervical canal stenosis below C3 also received C1 laminectomy and expansion laminoplasty between C3 and C6. Following surgery, a Philadelphia collar was postoperatively used for all patients for 1 month.

Assessment of neurologic and radiographic characteristics

The JOA score was used to evaluate cervical myelopathy. The authors evaluated surgical outcomes using the Hirabayashi [15] recovery rate [(postoperative JOA score – preoperative JOA score)/preoperative JOA score].

Radiographic assessment by conventional radiography and MRI to detect pseudotumor regression and postoperative instability of the cervical spine was done every 3 months. The maximum thickness of the retro-odontoid pseudotumor on sagittal T2 weighted MRI images was used to evaluate regression of the pseudotumor. The reduction ratio of each pseudotumor was calculated as: [(maximum preoperative thickness - maximum postoperative thickness)/maximum preoperative thickness]  $\times$  100. For this study, pseudotumors were divided into two groups based on pseudotumor regression: "unchanged" pseudotumors were those defined as having a reduction ratio <49 %, while "reduced" pseudotumors were those defined as having a reduction ratio >50 %, the level at which an apparent subarachnoid space is seen on postoperative MRI. To assess atlantoaxial instability, the atlas-dens interval (ADI) was measured on pre- and postoperative flexionextension radiographs. Atlantoaxial subluxation was defined as an ADI >4 mm at flexion, according to the criteria of White and Panjabi [16]. Cervical flexibility and alignment were also investigated to measure the O-C2 angle, defined as the angle between McGregor's line and the inferior endplate line in the C2 and C2-7 angle, measured between the inferior endplate of C2 and C7. The angle difference for each angle was defined as: postoperative angle at neutral position - preoperative angle at neutral position. A minus value indicates a kyphotic change and a plus value indicates a lordotic change. The range of motion (ROM) for each angle was defined as: the angle at extension - the angle at flexion. The difference in ROM for each angle was defined as: postoperative ROM preoperative ROM [17]. Radiographic measurements were performed independently by three spine surgeons using

institutional digital radiography software (Centricity Enterprise Web ver.3.0; GE Yokogawa Medical Systems, Tokyo, Japan). Results were averaged among the three reviewers.

# Results

Preoperative radiographic characteristics of the cervical spine

On examination of preoperative radiographic characteristics (Tables 1, 2), the mean  $(\pm SD)$  preoperative ADI at flexion was  $2.9 \pm 0.7$  mm (range 2.2–3.9 mm); no patient had atlantoaxial subluxation (Table 2). Computerized tomography (CT) revealed that one patient had OALL and one had spontaneous fusion at C3/4, two patients had OPLL, and three patients showed cervical kyphosis of less than -5 degrees of the C2–7 angle (Table 1). Therefore, five of seven patients had abnormal findings in the cervical spine, including OALL, OPLL, segmental fusion and kyphotic change. The preoperative mean  $(\pm SD)$  angles were  $36.2^{\circ} \pm 2.6^{\circ}$  (range 20.8°–46.5°) for O-C2 and  $-1.9^{\circ} \pm$  $0.9^{\circ}$  (range  $-13.5^{\circ}$  to  $9.8^{\circ}$ ) for C2-7 (Table 1). The preoperative mean ( $\pm$ SD) ROMs were 19.9  $\pm$  8.4° (range  $4.7^{\circ}-31.0^{\circ}$ ) for O-C2 and  $23.8^{\circ} \pm 12.5^{\circ}$  (range  $9.7^{\circ}-42.0^{\circ}$ ) for C2–7 ROM (Table 1).

Surgical outcome of C1 laminectomy (JOA score and radiographic analysis)

The mean ( $\pm$ SD) preoperative JOA score was 7.2  $\pm$  3.2/17 (range 2.5–12/17) and that for the postoperative JOA score was 14.1  $\pm$  2.6 (range 9.5–17) at final follow up (Table 2).

The progressive cervical myelopathy that had been recognized in all patients before surgery began to improve 1 month following surgery. The mean ( $\pm$ SD) Hirabayashi recovery rate was 71.0  $\pm$  23.5 % (range 40.0–100 %) (Table 2). There were no complications in any patient during the postoperative course. Pre- and postoperative JOA scores and the recovery rate did not show any statistically significant relationship with any of the radiographic measurements, including cervical alignment, ROM and size of pseudotumor.

From the radiographic analysis, the ADI (mean  $\pm$  SD) at flexion was  $2.9 \pm 0.7$  mm (range 2.2–3.9 mm) preoperatively, and  $2.9 \pm 0.8$  mm (2.1–3.9 mm) postoperatively (Table 2). The preoperative ADI at extension was  $2.6 \pm 0.6$ mm (range 1.1-3.7 mm) and the postoperative ADI at extension was 2.6  $\pm$  0.9 mm (range 1.2–3.7 mm) at final follow up (Table 2). The mean ( $\pm$ SD) angle measurements for the O-C2 angle were  $36.2^{\circ} \pm 2.6^{\circ}$  (range 20.8°-46.5°) (Table 1) preoperatively and  $33.0^{\circ} \pm 2.0^{\circ}$  (range  $18.3^{\circ}-37.2^{\circ}$ ) (Table 3) postoperatively, while those for the C2-7 angle decreased from the  $-1.9^{\circ} \pm 0.9^{\circ}$  (range  $-13.5^{\circ}$  to  $+9.8^{\circ}$ ) (Table 1) seen preoperatively to the  $-5.8^{\circ} \pm 6.2^{\circ}$  seen postoperatively (range  $-17.3^{\circ}$  to  $-0.3^{\circ}$ ) (Table 3). The C2–7 angle was reduced in all patients following surgery. The mean  $(\pm SD)$  postoperative O-C2 ROM was  $16.1^{\circ} \pm 6.4^{\circ}$  (range 5.0°–21.7°) while that of C2-7 was  $24.6^{\circ} \pm 16.5^{\circ}$  (range  $9.3^{\circ}$ -52.3°) (Table 3). In addition, differences between pre- and postoperative ROM measurements were  $-3.8^{\circ} \pm 3.3^{\circ}$  (range  $-13.7^{\circ}$  to  $+4.0^{\circ}$ ) for O-C2 and  $+0.9^{\circ} \pm 4.8^{\circ}$  (range  $-9.0^{\circ}$  to  $+10.3^{\circ}$ ) for C2-7 (Table 3). Thus, this surgery was seen to produce a slight kyphotic change in O-C2 and C2-7. O-C2 ROM was disturbed following surgery. However, additional atlantoaxial subluxation was not seen.

 Table 1
 Patient demographics and preoperative radiographic characteristics

Preoperative radiographic characteristics of the cervical spine								
Patient no.	Age (years)/sex	Symptoms	OALL/OPLL	Kyphosis	Angle (°)		ROM (°)	
					O-C2	C2-7	O-C2	C2–7
1	76/M	Myelopathy/axial pain	-	-	$20.8\pm1.8$	$6.3\pm0.6$	31.0 ± 1.7	$25.3\pm5.8$
2	64/M	Myelopathy	_	-	$36.7\pm1.0$	$9.8\pm0.3$	$16.0\pm6.6$	$40.3\pm3.5$
3	73/M	Myelopathy/axial pain	_	+	$46.5\pm3.0$	$-7.3\pm1.3$	$22.3\pm3.2$	$15.3\pm3.5$
4	87/M	Myelopathy	C3-7 OPLL	+	$39.7\pm3.8$	$-13.5 \pm 1.7$	$20.0 \pm 1.0$	$18.3 \pm 1.2$
5	70/M	Myelopathy	C3/4 fusion	-	$35.0\pm2.2$	$-2.3\pm0.6$	$19.3\pm2.5$	$42.0\pm4.0$
6	82/M	Myelopathy	C3-6 OALL	+	$37.5\pm3.5$	$-6.0\pm1.3$	$25.7\pm5.0$	$15.3\pm2.1$
7	77/F	Myelopathy/axial pain	C3-6 OPLL	-	$36.7\pm2.8$	$-0.5\pm0.9$	$4.7 \pm 1.5$	$9.7\pm3.5$
Mean	$75.6\pm7.6$				$36.2\pm2.6$	$-1.9\pm0.9$	$19.9\pm8.4$	$23.8\pm12.5$

Data is shown as mean  $\pm$  standard deviation. Kyphosis was defined as less than  $-5^{\circ}$  of the C2–7 angle

OALL ossification of the anterior longitudinal ligament, OPLL ossification of the posterior longitudinal ligament, ROM range of motion

#### Eur Spine J (2013) 22:1119-1126

Outcomes of surgical treatment								
Patient no.	Surgery	JOA score (full score: 17)			Preoperative ADI (mm)		Postoperative ADI (mm)	
		Preoperative	Postoperative	Recovery rate (%)	Flexion	Extension	Flexion	Extension
1	C1 laminectomy	4	15	84.6	$2.3\pm0.5$	$1.8 \pm 0.3$	$3.9\pm0.1$	$1.8 \pm 0.3$
2	C1 laminectomy	12	17	100	$2.2\pm0.4$	$2.3\pm0.2$	$2.3\pm0.3$	$2.3\pm0.2$
3	C1 laminectomy + C3–6 laminoplasty	8	14.5	72.2	3.9 ± 0.2	3.7 ± 0.3	2.1 ± 0.2	$2.2 \pm 0.3$
4	C1 laminectomy	5.5	12	56.5	$3.1\pm0.3$	$1.1\pm0.2$	$2.3\pm0.2$	$1.2\pm0.2$
5	C1 laminectomy	6.5	16.5	95.2	$3.8\pm0.1$	$3.7\pm0.3$	$3.8\pm0.2$	$3.7\pm0.1$
6	C1 laminectomy + C3–6 laminoplasty	2.5	9.5	48.3	$2.5\pm0.3$	2.4 ± 0.3	$2.2\pm0.3$	2.1 ± 0.2
7	C1 laminectomy + C3–6 laminoplasty	12	14	40.0	2.9 ± 0.2	3.0 ± 0.5	$3.2 \pm 0.5$	3.0 ± 0.5
Mean		$7.2 \pm 3.2$	$14.1\pm2.6$	$71\pm23.5$	$2.9\pm0.7$	$2.6\pm0.6$	$2.9\pm0.8$	$2.6\pm0.9$

# Table 2 Surgical treatment and outcomes

Data is shown as mean  $\pm$  standard deviation

JOA Japanese Orthopaedic Association, ADI atlas-dens interval

Table 3 Postoperative radiographic characteristics

Radiographic characteristics of O-C2 and C2-7								
Patient no.	0-C2 (°)				C2–7 (°)			
	Postoperative angle	Angle difference	Postoperative ROM	ROM difference	Postoperative angle	Angle difference	Postoperative ROM	ROM difference
1	$18.3\pm0.8$	$-2.5\pm2.5$	$17.3\pm2.5$	$-13.7 \pm 1.5$	$-2.7 \pm 1.2$	$-9.0\pm1.0$	19.3 ± 1.2	$-6.0 \pm 5.3$
2	$37.0\pm2.6$	$0.3 \pm 3.1$	$20.0\pm2.6$	$+4.0\pm6.9$	$-2.7\pm0.3$	$-12.5\pm0.5$	$45.3\pm3.5$	$+5.0\pm7.0$
3	$37.2\pm2.0$	$-9.3\pm2.1$	$15.7 \pm 2.1$	$-6.7\pm1.2$	$-17.3 \pm 1.0$	$-10.0\pm1.0$	$12.7\pm0.6$	$-2.7 \pm 3.1$
4	$32.5\pm2.3$	$-7.2\pm1.5$	$21.7 \pm 2.5$	$+1.7 \pm 3.1$	$-12.3 \pm 1.8$	$1.2 \pm 3.4$	$9.3\pm5.5$	$-9.0 \pm 5.6$
5	$31.9\pm3.4$	$-3.1 \pm 1.3$	$11.5 \pm 2.6$	$-7.8\pm3.8$	$-2.2 \pm 2.0$	$0.2\pm2.4$	$52.3 \pm 1.5$	$+10.3 \pm 4.7$
6	$37.0\pm1.0$	$-0.5\pm2.6$	$21.3\pm5.0$	$-4.3 \pm 1.2$	$-4.0\pm0.5$	$2.0 \pm 1.8$	$21.3\pm3.8$	$+6.0 \pm 5.6$
7	$36.8\pm2.1$	$0.2 \pm 1.4$	$5.0\pm5.3$	$+0.3\pm5.5$	$-0.3\pm0.3$	$0.8\pm0.6$	$12.0\pm1.7$	$+2.3\pm2.3$
Mean	$33.0\pm2.0$	$-3.2\pm2.1$	$16.1\pm6.4$	$-3.8\pm3.3$	$-5.8\pm6.2$	$-3.9\pm1.7$	$24.6\pm16.5$	$+0.9\pm4.8$

Data is shown as mean  $\pm$  SD. Angle difference (°) = Angle (postoperative) – Angle (preoperative). ROM difference (°) = ROM (postoperative) – ROM (preoperative)

ROM range of motion

# Reduction of retro-odontoid pseudotumor

In all patients, the pseudotumors began to reduce within 12 months following surgery. The mean ( $\pm$ SD) reduction rate was 54.6  $\pm$  26.6 % (range 23.0–78.9 %). Four patients had more than 70 % reduction (Table 4); their pseudotumors were classified as reduced, a clinically complete reduction (Figs. 1, 2). These reduced pseudotumors began to reduce faster than the unchanged pseudotumors. The reduction process plateaued at almost 1 year after surgery. No recurrence or regrowth occurred during the follow up period in any patient. The duration of tumor

regression did not correlate with tumor size, radiographic measurements, including OALL, OPLL, segmental fusion or kyphotic change. Additionally, the reduction rate did not show any statistical significance with JOA score or recovery rate.

From MRI analysis after Gd administration in the five patients who underwent this procedure, the pseudotumor was reduced in the four patients who had shown Gd enhancement of the tumor (Table 4). In contrast, in the one patient who showed no Gd enhancement, the pseudotumor did not reduce and the reduction rate remained at 25 %.

Table 4	Postoperative	reduction	of	retro-odontoid	pseudotumor
---------	---------------	-----------	----	----------------	-------------

Reduction of retro-odontoid pseudotumor							
Patient no.	Preoperative MI	RI	Follow up MRI (reduction rate, %				
	T1	T2	Gd enhance				
1	Low	Low	Enhance	Reduction (76.9)			
2	Low	Iso/high	Enhance	Reduction (72.2)			
3	Low/iso	Iso/high	Enhance	Reduction (75.0)			
4	Low	Iso/high	Enhance	Reduction (78.9)			
5	Low	Low	-	Non reduction (23.0)			
6	Low/iso	Low	-	Non reduction (31.0)			
7	Low/iso	Low	Non enhance	Non reduction (25.0)			



Fig. 1 Patient 1: 76 year-old man. The retro-odontoid pseudotumor was revealed by sagittal (a) and axial (c) images on T2-weighted MRI. Capsular enhancement is shown by Gd administration MRI (b).

# Discussion

Retro-odontoid pseudotumors are uncommon and often hard to diagnose clinically because clinical symptoms and signs vary and include paraparesis, headache, neck stiffness and nuchal pain [18]. Pseudotumors occasionally result in progressive myelopathy or sudden death. The most frequent cause of retro-odontoid pseudotumor is the pannus of inflammatory granulation tissue arising from the synovium around the dens in rheumatoid arthritis patients. In fact, retro-odontoid pseudotumor is

Following C1 laminectomy, the pseudotumor is reduced to 70 % at 412 days ( $\mathbf{e}$ ,  $\mathbf{f}$ ). Comparing pre- and postoperative radiographs ( $\mathbf{d}$ ,  $\mathbf{g}$ ), additional atlantoaxial subluxation is not seen

often recognized in rheumatoid arthritis. Pettersson et al. [19] analyzed cervical spine MR images of 23 rheumatoid arthritis patients and found that 14 had periodontoid pannus, all with atlantoaxial subluxation. They concluded that the large soft-tissue mass is the sequela of chronic mechanical irritation caused by atlantoaxial subluxation. In contrast, an atypical mass at the posterior of the odontoid process not associated with underlying disease has been reported to be a reactive fibrocartilaginous mass induced by chronic subluxation in the craniovertebral junction [5, 6]. The pathomechanism for pseudotumor

![](_page_5_Figure_2.jpeg)

Fig. 2 Patient 6: 82 year-old man. The retro-odontoid pseudotumor is shown by sagittal (a) and axial (b) images on T2-weighted MRI. Following C1 laminectomy, the pseudotumor remains at a reduction

formation has been postulated to the result of atlantoaxial instability that induces repeated tears and subsequent hypertrophy of the transverse ligaments [8, 20]. Chikuda et al. [10] found that retro-odontoid pseudotumor was not necessarily associated with overt atlantoaxial instability, but that excessive stress concentration to the atlantoaxial complex caused by altered biomechanics of the cervical spine due to OALL, OPLL and cervical kyphosis may cause repeated damage to the transverse ligament. In our study no patient had atlantoaxial subluxation. However, five of the seven patients had risk factors which Chikuda et al. [10] reported, ankylosis of the segments adjacent to the atlantoaxial joint and cervical kyphosis below the atlantoaxial joint. The results of our study support Chikuda's hypothesis.

A single preferred therapeutic strategy for retro-odontoid pseudotumor has not been established. Crockard et al. [6] directly resected the pseudotumor using a transoral approach. Because previous reports identified retro-odontoid pseudotumor as a reactive mass caused by mechanical stress due to atlantoaxial instability or ankylosis in the mid and lower cervical spine, posterior fusion, such as O-C2 fusion or O-C4 fusion, has been considered to be the appropriate procedure for preventing further progression of the pathology. Additionally, posterior fusion resulted in regression of the retro-odontoid pseudotumor and improvement of neurological deficits [8, 10].

rate of 31.3 % ( $\mathbf{d}$ ,  $\mathbf{e}$ ). Comparing pre- and postoperative radiographs ( $\mathbf{c}$ ,  $\mathbf{f}$ ), additional atlantoaxial instability is not seen

In contrast, Suetsuna et al. [14] reported that two of three retro-odontoid pseudotumor patients had spontaneous regression after C1 laminoplasty using hydroxyapatite. In terms of reduction rate, the authors considered that C1 laminectomy seemed inferior to posterior fusion. Our study, however, showed that C1 laminectomy resulted in spontaneous regression of pseudotumors and satisfactory improvement of neurological deficit in all patients. Furthermore, additional atlantoaxial instability was not observed during the follow up period, which exceeded 2 years.

The craniocervical junction (CCJ) is composed of three types of components, bone (occipital bone, atlas and axis), joint, and accompanying ligament. The CCJ protects the upper cervical cord, brain stem and lower cranial nerves, and simultaneously provides an exquisite structure to enable excellent flexibility in the O-C1 and C1-2 joints. Specifically, the O-C1 joint represents 50 % of the total flexextension motion in the entire cervical spine, while the C1-2 joint accounts for 50 % of the rotational motion [16]. The mechanical stability of the CCJ is mainly provided by ligaments. The transverse and alar ligaments are particularly important for controlling the CCJ, and these ligaments attach to the posterior odontoid process [21]. The posterior occipitoatlantal membrane and posterior atlantoaxial membrane are attached to the posterior arch of the atlas; however, these membranes are of less importance than the transverse and alar ligaments for controlling the CCJ.

While retro-odontoid pseudotumor without atlantoaxial instability is often associated with OALL, OPLL and severe spondylosis, the cervical ROM of retro-odontoid pseudotumor patients was originally disturbed. Posterior fusion would lead to further ROM disturbance, so should be avoided. Based on the anatomical properties and the results of this study, C1 laminectomy is not considered to induce postoperative atlantoaxial instability, and is, therefore, a candidate for surgical procedure for treating pseudotumor without atlantoaxial instability. However, longer term follow up is needed to assess potential instability of the upper cervical spine.

Following treatment for retro-odontoid pseudotumor, regression may occur. Spontaneous regression of herniated nucleus pulposus (NP) is a clinically demonstrated phenomenon in intervertebral disc herniation. Recent advances in imaging techniques, CT and MRI, have facilitated the precise documentation of this process [22]. Slavin et al. hypothesized [23] that cartilaginous tissue resorption results from enzymatic degradation and phagocytosis as a result of inflammation and neovascularization due to an autoimmune response [24, 25]. Some studies have revealed neovascularization at the periphery of the sequestrated disc and the presence of macrophages and T cells in the vascularized areas [26–28]. In our study, all patients with reduced pseudotumors demonstrated contrast enhancement of their pseudotumor on MRI after Gd administration. This enhancement is considered to reflect neovascularization around the retroodontoid pseudotumor. Although retro-odontoid pseudotumor and herniated nucleus pulposus are different pathologies, the authors of this study suggest that the improvement of blood flow in the neovascularized area resulting from C1 laminectomy may contribute to the regression of the retro-odontoid pseudotumor.

The best therapeutic strategy for treating retro-odontoid pseudotumor patients with atlantoaxial subluxation should be posterior fusion. Because patients without atlantoaxial subluxation, especially those showing enhancement on MRI after Gd administration, are predisposed to spontaneous regression after C1 laminectomy, we consider this procedure best for treatment of retro-odontoid pseudotumor patients without atlantoaxial subluxation. Furthermore, even for those patients who show no Gd enhancement, C1 laminectomy may be an appropriate surgery for patients without atlantoaxial subluxation, because our study found satisfactory neurological improvement and no additional instability following C1 laminectomy.

# Conclusion

C1 laminectomy for treatment of retro-odontoid pseudotumor in the absence of atlantoaxial instability improved neurological deficits in all patients. Additional atlantoaxial instability was not observed. All pseudotumors were reduced after C1 laminectomy, four patients who showed enhancement on MRI after Gd administration had their tumors almost completely resolved after 1 year. Therefore, C1 laminectomy was considered the therapeutic option for retro-odontoid pseudotumor without atlantoaxial subluxation in addition to C1/2 fixation.

Conflict of interest None.

#### References

- Grob D, Wursch R, Grauer W, Sturzenegger J, Dvorak J (1997) Atlantoaxial fusion and retrodental pannus in rheumatoid arthritis. Spine (Phila Pa 1976) 22:1580–1583 (discussion 1584)
- Kenez J, Turoczy L, Barsi P, Veres R (1993) Retro-odontoid "ghost" pseudotumours in atlanto-axial instability caused by rheumatoid arthritis. Neuroradiology 35:367–369
- Hatakeyama A, Fujinaga H, Togo T, Tamura T, Ozawa K, Shoji T, Sasaoka T (1992) Remarkable improvement of activity by CAPD in a hemodialysis patient with a pseudotumor of the craniocervical junction. Adv Perit Dial 8:116–119
- Rousselin B, Helenon O, Zingraff J, Delons S, Drueke T, Bardin T, Moreau JF (1990) Pseudotumor of the craniocervical junction during long-term hemodialysis. Arthritis Rheum 33: 1567–1573
- Sze G, Brant-Zawadzki MN, Wilson CR, Norman D, Newton TH (1986) Pseudotumor of the craniovertebral junction associated with chronic subluxation: MR imaging studies. Radiology 161:391–394
- Crockard HA, Sett P, Geddes JF, Stevens JM, Kendall BE, Pringle JA (1991) Damaged ligaments at the craniocervical junction presenting as an extradural tumour: a differential diagnosis in the elderly. J Neurol Neurosurg Psychiatry 54:817–821
- Jun BY, Yoon KJ, Crockard A (2002) Retro-odontoid pseudotumor in diffuse idiopathic skeletal hyperostosis. Spine (Phila Pa 1976) 27:E266–E270
- Yoshida K, Hanyu T, Takahashi HE (1999) Progression of rheumatoid arthritis of the cervical spine: radiographic and clinical evaluation. J Orthop Sci 4:399–406 pii:90040399.776
- Tanaka S, Nakada M, Hayashi Y, Mohri M, Uchiyama N, Hamada J Retro-odontoid pseudotumor without atlantoaxial subluxation. J Clin Neurosci 17:649–652. doi:10.1016/j.jocn.2009.07.116
- Chikuda H, Seichi A, Takeshita K, Shoda N, Ono T, Matsudaira K, Kawaguchi H, Nakamura K (2009) Radiographic analysis of the cervical spine in patients with retro-odontoid pseudotumors. Spine (Phila Pa 1976) 34:E110–E114. doi:10.1097/BRS.0b013e31818 acd27
- Isono M, Ishii K, Kamida T, Fujiki M, Goda M, Kobayashi H (2001) Retro-odontoid soft tissue mass associated with atlantoaxial subluxation in an elderly patient: a case report. Surg Neurol 55:223–227 pii:S0090-3019(01)00345-7
- Cihanek M, Fuentes S, Metellus P, Pech-Gourg G, Dufour H, Grisoli F (2008) Disappearance of retro-odontoid pseudotumor after C1–C2 transarticular fixation screw. Neurochirurgie 54:32– 36. doi:10.1016/j.neuchi.2007.12.002
- Yamaguchi I, Shibuya S, Arima N, Oka S, Kanda Y, Yamamoto T (2006) Remarkable reduction or disappearance of retroodontoid pseudotumors after occipitocervical fusion. Report of three cases. J Neurosurg Spine 5:156–160. doi:10.3171/spi.2006.5.2.156
- Suetsuna F, Narita H, Ono A, Ohishi H (2006) Regression of retroodontoid pseudotumors following C-1 laminoplasty. Report of three cases. J Neurosurg Spine 5:455–460. doi:10.3171/spi. 2006.5.5.455

- 15. Hirabayashi K, Miyakawa J, Satomi K, Maruyama T, Wakano K (1981) Operative results and postoperative progression of ossification among patients with ossification of cervical posterior longitudinal ligament. Spine (Phila Pa 1976) 6:354–364
- 16. White AI, Panjabi MM (1978) Clinical biomechanics of the spine. JB Lippincott, Philadelphia
- Miyata M, Neo M, Fujibayashi S, Ito H, Takemoto M, Nakamura T (2009) O-C2 angle as a predictor of dyspnea and/or dysphagia after occipitocervical fusion. Spine (Phila Pa 1976) 34:184–188. doi:10.1097/BRS.0b013e31818ff64e
- Liccardo G, Lunardi P, Menniti A, Floris R, Pastore FS, Fraioli B (2003) Calcifying pseudo-tumor of the spine: description of a case and review of the literature. Eur Spine J 12:548–551. doi:10.1007/s00586-003-0546-7
- Pettersson H, Larsson EM, Holtas S, Cronqvist S, Egund N, Zygmunt S, Brattstrom H (1988) MR imaging of the cervical spine in rheumatoid arthritis. Am J Neuroradiol 9:573–577
- Matsumoto T, Takada S, Tsujimoto K, Ozaki T, Ishimoto K, Tsumura N, Shiba R, Kurosaka M (2006) Enlarging retro-odontoid pseudotumor after expanding cervical laminoplasty in the presence of kyphosis. Spine J 6:228–232. doi:10.1016/j.spinee. 2005.08.010
- Werne S (1957) Studies in spontaneous atlas dislocation. Acta Orthop Scand Suppl 23:1–150

- 22. Sei A, Nakamura T, Fukuyama S, Ikeda T, Senda H, Takagi K (1994) Spontaneous regression of lumbar hernia of the nucleus pulposus. Follow-up study of 4 cases by repeated magnetic resonance imaging. Rev Chir Orthop Reparatrice Appar Mot 80: 144–149 pii:78241
- Slavin KV, Raja A, Thornton J, Wagner FC Jr (2001) Spontaneous regression of a large lumbar disc herniation: report of an illustrative case. Surg Neurol 56:333–336. pii:S0090-3019(01) 00607-3 (discussion 337)
- 24. Doita M, Kanatani T, Harada T, Mizuno K (1996) Immunohistologic study of the ruptured intervertebral disc of the lumbar spine. Spine (Phila Pa 1976) 21:235–241
- Saal JA (1996) Natural history and nonoperative treatment of lumbar disc herniation. Spine (Phila Pa 1976) 21:2S–9S
- 26. Ito T, Yamada M, Ikuta F, Fukuda T, Hoshi SI, Kawaji Y, Uchiyama S, Homma T, Takahashi HE (1996) Histologic evidence of absorption of sequestration-type herniated disc. Spine (Phila Pa 1976) 21:230–234
- Woertgen C, Rothoerl RD, Brawanski A (2000) Influence of macrophage infiltration of herniated lumbar disc tissue on outcome after lumbar disc surgery. Spine (Phila Pa 1976) 25:871–875
- Arai Y, Yasuma T, Shitoto K, Yamauchi Y, Suzuki F (2000) Immunohistological study of intervertebral disc herniation of lumbar spine. J Orthop Sci 5:229–231. doi:10.1007/s007760000050229.776