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치상돌기 후방부 가성 종양의 후방 고정 수술 후 퇴행

Postoperative regression of retro-odontoid pseudotumor after atlantoaxial posterior fixation

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Abstract

Postoperative regression of retro-odontoid pseudotumor after atlantoaxial posterior fixation

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Purpose: To investigate the incidence of retro-odontoid pseudotumor in patients with atlantoaxial instability (AAI) and evaluate pseudotumor regression after posterior fixation.

Materials and Methods: From July 2004 to August 2015, 175 patients with AAI underwent posterior fixation operations at our institution. After excluding 11 patients (previous operation, n = 4; history of tumor, n = 7), the final study population comprised 164 patients. The final study population was categorized according to their underlying diseases (rheumatoid arthritis [RA], os odontoideum, atlanto-occipital assimilation, dens fracture, AAI of unknown cause, etc.) and age (adult and pediatric groups). The incidence of retro-odontoid pseudotumor in each group was analyzed. Pre- and postoperative magnetic resonance or computed tomography images were reviewed to assess its regression following surgery.

Results: Of the 164 patients included, 38 had retro-odontoid pseudotumor (23.2%). Three were diagnosed with RA and the rest were non-RA patients including os odontoideum (n = 12), dens fracture (n = 6), atlanto-occipital assimilation (n = 4), Morquio syndrome (n = 1), and AAI of unknown cause (n = 12). Pseudotumor size regressed in all 38 patients after atlantoaxial posterior fixation. There was a statistically significant decrease in pseudotumor size (the length between the anterior border of the odontoid process to the posterior border of the pseudotumor) from a mean length of 17.7 mm to 14.9 mm ($P < 0.001$).

Conclusions: The patients had various underlying diseases and the overall incidence of retro-odontoid pseudotumor in patients with symptomatic AAI was 23.2% at our institution during the past 11 years. All patients who underwent posterior fixation for AAI showed a statistically significant decrease in pseudotumor size.

Keywords: Retro-odontoid pseudotumor, Atlantoaxial instability, Posterior fixation, Magnetic resonance imaging, Computed tomography

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Table of contents

List of tables

List of figures

Introduction

Retro-odontoid pseudotumors or peri-odontoid pseudotumors are mass-like lesions involving the odontoid process and its surrounding structures, including the anterior arch of the atlas and transverse ligaments.[1] Many reports have shown associations between retro-odontoid pseudotumor and various diseases such as rheumatoid arthritis (RA), os odontoideum, odontoid fracture, atlas hypoplasia, hemodialysis and even chronic atlantoaxial instability (AAI) of unknown cause.[2-4] RA is a well-known disease that can form a retro-odontoid pseudotumor, more often called inflammatory pannus, and many previous studies reported that the pseudotumor resulted from a combination of chronic AAI and an inflammatory process.[1]

Chronic AAI caused by various etiologies is postulated to be one of the underlying pathophysiologies of pseudotumor formation. Chronic AAI may cause compression of the upper cervical cord and even the medulla oblongata, and retro-odontoid pseudotumor formation in chronic AAI patients may aggravate cord compression and accelerate neurologic deterioration if not treated in time.[5,6]

Recently, many studies suggested that posterior fixation alone is sufficient for the reduction of a retro-odontoid pseudotumor, as opposed to anterior (trans-oral/trans-pharyngeal) surgical approaches.[1,6-9] However, regression of retro-odontoid pseudotumor after posterior fixation has been reported more frequently in patients with RA, and there is no large case study encompassing both RA and non-RA patients. Moreover, the incidence of retro-odontoid pseudotumors in AAI patients has not yet been reported.

Thus, the purpose of our study was to investigate the incidence of retro-odontoid pseudotumors in patients with AAI and to evaluate pseudotumor regression after posterior fixation.

Materials and Methods

The institutional review board approved the study protocol and waived the requirement for informed consent for this retrospective study.

Study Population

From July 2004 to August 2015, 175 patients with AAI underwent posterior fixation operations at Seoul National University Bundang Hospital. Electronic medical records (EMRs) and a picture archiving and communication system (PACS) were used to retrieve clinical information about the patients, including operation records, radiologic reports, and computed tomography (CT) and magnetic resonance (MR) images. The final study population comprised 164 patients after excluding 11 patients who underwent a previous operation (n = 4) or who had a history of tumor (n = 7) or infection (n = 0) at the cervical spine. Patients included in the final study population were categorized according to their underlying diseases (RA, os odontoideum, atlanto-occipital assimilation, dens fracture, AAI of unknown cause, etc.) and the incidence proportion of retro-odontoid pseudotumor in each symptom complex or diagnosis was analyzed. Furthermore, study population was additionally categorized into adult and pediatric patients. The incidence proportion of retro-odontoid pseudotumor in each age group was also analyzed.

Image Acquisition

All patients underwent CT using a 64-slice CT scanner (Brilliance-64, Philips Medical Systems, Best, The Netherlands) or a 256-slice CT scanner (Brilliance iCT, Philips Medical Systems). Scanning parameters were as follows: 120 kV; 250 mAs; collimation, 64 × 0.625 mm (64-slice CT) or 128 × 0.625 mm (256-slice CT); rotation time, 0.5 s; volume CT dose index, 16.40 mGy (64-slice CT) or 16.951 mGy (256-slice CT); and dose-length product, 520.1 mGy · cm (64-slice CT) or 602.81 mGy · cm (256-slice CT). The sagittal plane was reconstructed with a slice thickness and slice interval of 3 mm each.

Most patients underwent MR imaging (MRI) at our institution using a 1.5-T scanner (Gyroscan Intera, Philips Medical Systems) with a Synergy Spine Coil (Philips Medical Systems) or a 3.0-T scanner (Gyroscan Intera Achieva, Philips Medical Systems). Patients were placed in the supine position with a cushion under both knees. Sagittal and axial T1-weighted (T1W) spin-echo and T2-weighted (T2W) fast spin-echo images were obtained using the following parameters: repetition time/echo time, 500/15 msec (T1W images) or 3600/120 msec (T2W images); slice thickness, 4 mm; slice gap, 0.4 mm; field-of-view, 32 cm (sagittal images) or 16 cm (axial images); matrix, 512 × 512; flip angle, 90° ; and excitations, 3. Some had preoperative MRIs imported from outside hospitals obtained on various 1.5-T scanners. All outside images included sagittal T1W and T2W images with comparable image quality.

Image Interpretation

Two readers (E.L. [4 years of experience in spine radiology] and J.H.P. [third year resident in radiology]) were blinded to the clinical findings and reviewed all 164 patients' preoperative MR or CT images to evaluate the presence or absence of retro-odontoid pseudotumor. In the second interpretation session, the two readers resolved discrepancies in consensus and determined the presence of pseudotumor. In the third interpretation session, pre- and postoperative CT and MR images were randomly analyzed by both readers, and the size of the retro-odontoid pseudotumor was measured. The size of the pseudotumor was measured in its largest sagittal diameter, from the anterior border of the odontoid process to the posterior border of the pseudotumor (Figure 1). The latest follow-up image was chosen for postoperative image evaluation. The size difference between pre- and postoperative MR or CT images was compared using the mean of the two measured values from both readers.

Postoperative regression percentage of the retro-odontoid pseudotumor was calculated considering only the posteriorly bulging portion of the pseudotumor. The measurement was performed in sagittal images, from the posterior border of the odontoid process to the posterior border of the pseudotumor. The size difference in preoperative and postoperative images was used to calculate the regression percentage, and consensus was reached between the two readers.

Degree of preoperative cervical central canal compromise was evaluated using the new MRI grading system for the cervical canal stenosis.[10] Cervical canal stenosis was classified according to the T2-weighted sagittal images into the following grades: grade 0, absence of canal stenosis; grade 1, subarachnoid space obliteration exceeding 50%; grade 2, spinal cord deformity; and grade 3, spinal cord signal change.

All patients in the study population had preoperative MR and CT images but some of them had been evaluated only via CT post-operatively. For patients who had postoperative CT images only, measurement of retro−odontoid pseudotumor size was performed using preoperative and postoperative CT images. For patients who had both postoperative CT and MR images, measurement was performed using MR images only.

Clinical Data Assessment

Preoperative and postoperative clinical data assessment was done by reviewing orthopedic surgeon ' s clinical data assessment sheet. Neck Disability Index (NDI) [11] percentage and Japanese Orthopedic Assessment (JOA) score[12] was chosen for comparison of disease severity scores before and after posterior fixation. All patients with retro−odontoid pseudotumor had preoperative and postoperative NDI percentage. However 6 patients were lack of preoperative or postoperative JOA scores. Therefore, preoperative and postoperative NDI percentages of 38 patients and JOA scores of 32 patients were compared to evaluate changes in disease severity scores. Postoperative 6 months score was chosen for the postoperative disease severity score.

The recovery rate of JOA score was calculated using the Hirabayashi method[13] as follows: (postoperative score − preoperative score)/(17 − preoperative score) x 100. Calculated recovery rates were classified into five groups: excellent (75 % < to ≤ 100 %), good (50 % < to ≤ 75 %), fair (25 % < to ≤ 50 %), unchanged (0 % < to ≤ 25 %), and worse (< 0 %).[14]

Statistical Analysis

A paired *t*−test was used to compare the pre− and postoperative sizes of the retro−odontoid pseudotumor. Wilcoxon's signed−rank test was used to investigate size differences in each symptom complex or diagnosis group. Statistical significance was set as $P \leq 0.05$.

Results

Among the total of 164 patients included in our study population (104 females; age range, 3–76 years; mean age, 47 years), 38 patients had retro-odontoid pseudotumor (25 females; age range, 4–72 years; mean age, 57 years) at the preoperative evaluation. Of the 38 patients, three patients were diagnosed with RA and the rest were non-RA patients with diseases including os odontoideum (n = 12), dens fracture (n = 6), atlanto-occipital assimilation (n = 4), Morquio syndrome (n = 1), and AAI of unknown cause (n = 12) (Table 1).

Retro-odontoid Pseudotumor Regression

The size of the retro-odontoid pseudotumor regressed in all 38 patients after atlantoaxial posterior fixation (Table 2). There was a statistically significant interval decrease in the size of the pseudotumor from a mean length of 17.7 mm to 14.9 mm ($P < 0.001$) (Table 3). A statistically significant size decrease of the pseudotumor was noted in patients with AAI of unknown cause (Figure 2), os odontoideum (Figure 3) and dens fracture (Figure 4), while statistical significance was not evident in patients with atlanto-occipital assimilation or RA. The mean follow-up period was 12.5 months.

There was also statistically significant size decreased of the posteriorly bulging portion of the pseudotumor after surgery and the mean percentage of regression was 72.1% ($P < 0.001$) (Table 3). The percentage of regression after posterior fixation in each patient and in each symptom complex or diagnosis group is shown in Table 2 and Table 3 respectively.

The sagittal measurement of the retro-odontoid pseudotumor was performed using CT images in nine patients and MR images in 29 patients. All preoperative and postoperative CT images were acquired at our institute and three patients out of the 29 had preoperative MRI imported from outside hospitals.

Incidence Proportion

The incidence proportion (cumulative incidence) of retro-odontoid pseudotumor among patients with symptomatic AAI was 23.2% (adult patients, 24%; pediatric patients, 16.7%). The highest incidence proportion in adult patients was noted in the atlanto-occipital assimilation group (42.9%), followed by AAI of unknown cause (30.6%), dens fracture (26.1%), RA (21.4%), and os odontoideum (20.8%). Thirteen patients were classified as miscellaneous and included multiple C1-2 fractures other than dens fracture (n = 5), congenital block vertebra C1-2 (n = 1), basiooccipital hypoplasia (n = 1), pseudogout (n = 1), psoriatic arthritis (n = 1), diffuse idiopathic skeletal hyperostosis (DISH) (n = 1), Morquio syndrome (n = 1), Gorham's disease (n = 1), and asymmetrical collapse of the C1-2 facet joint (n = 1). One patient with Morquio syndrome in this group was positive of retro-odontoid pseudotumor. Three patients in pediatric population group had retro-odontoid pseudotumor (AAI of unknown cause, n = 1; os odontoideum, n = 1; atlanto-occipital assimilation, n = 1). The incidence proportion of retro-odontoid pseudotumor in pediatric patients within each symptom complex or diagnosis is described in Table 1.

Clinical Outcome

Among 38 patients with retro-odontoid pseudotumor 30 patients had signs and symptoms of myelopathy (79%), six patients complained of posterior neck pain (16%) and two patients had radiculopathy (5%). Regarding preoperative central canal compromise, 28 patients had compressive myelopathy on MRI and were classified as grade 3. Four patients were classified as grade 2 and six patients were classified as grade 1 (Table 2).

Postoperative disease severity scores showed statistically significant improvement in these patients. The mean JOA score improved from 11.3 (range 5.5–16.5) to 13.4 (range 5.5–17) at 6 months follow-up after surgery (P = 0.007). The mean NDI percentage also improved from 41.5 (range 4–76) to 28.3 (range 2–52) at 6 months follow-up ($P < 0.001$) (Table 4). Postoperative neurological recovery rate was excellent in 28.1% (n = 9), good in 12.5% (n = 4), fair in 15.6% (n = 5), unchanged in 25% (n = 8), and worse in 18.8% (n = 6) of patients (Table 5).

Among 38 patients with retro-odontoid pseudotumor, 33 patients had undergone atlantoaxial fusion and five patients had undergone occipitocervical fusion (O–C2, n = 3; O–C3, n = 1; O–C5, n = 1). Of the 38 patients, only two patients (5.3%) required re-operation. One patient with AAI of unknown cause (case no. 12) underwent trans-oral decompression after 6 months because the initial size of the retro-odontoid pseudotumor was very large and there was not enough regression to relieve compressive myelopathy. Another patient with underlying atlanto-occipital assimilation (case no. 32) underwent re-operation for hematoma evacuation 4 days after the first operation.

Retro-odontoid Pseudotumor Signal Intensity

The signal intensity (SI) of the retro-odontoid pseudotumor was lower than that of the spinal cord in both T1W and T2W images in the majority of cases (n = 24; AAI of unknown cause, 9; os odontoideum, 8; dens fracture, 4; atlanto-occipital assimilation, 3). Thirteen patients showed low T1 and mixed T2 SI (os odontoideum, 4; RA, 3; AAI of unknown cause, 2; atlanto-occipital assimilation, 2; dens fracture, 2), and one patient with AAI of unknown cause showed low T1 and high T2 SI.

Discussion

To our knowledge, this is the first study to demonstrate the incidence proportion of retro-odontoid pseudotumor in patients with symptomatic AAI. Our study showed that 23.2% (adult patients, 24%; pediatric patients, 16.7%) of patients with AAI had retro-odontoid pseudotumor, and this study population comprised various underlying diseases. The atlanto-occipital assimilation group had the highest incidence proportion (42.9%) in adult patients, but this seemingly high incidence proportion might have resulted from the small sample size in this symptom complex. Eleven out of 36 patients with AAI of unknown cause in adult group revealed to have retro-odontoid pseudotumor, resulting in a 30.6% incidence proportion. None of the patients in this group had significant underlying diseases such as diabetes mellitus or RA. Tojo et al. reported that retro-odontoid pseudotumor had positive correlations with age, degenerative changes of the cervical spine, and long-term dialysis.[15] The majority of AAI of unknown cause patients with retro-odontoid pseudotumor in this study were elderly.

However, none of them had a history of dialysis. Our study population included only 16 patients diagnosed with RA. Three among them showed retro-odontoid pseudotumor, resulting in an incidence proportion of 21.4%. The small size of the patient population in the RA group might be due to the small number of patients with severe RA visiting our hospital or partly due to advancement in medication resulting in good disease control. Our study included a large number of patients with os odontoideum and this disease group's incidence proportion of retro-odontoid pseudotumor was 20.8% in adult patients and 12.5% in pediatric patients.

Though the study population had a variety of underlying diseases, retro-odontoid pseudotumor showed regression in all patients, irrespective of the underlying disease. This was statistically significant when all 38 patients with retro-odontoid pseudotumor were considered as a whole. When each disease group was considered separately, statistical significance was not achieved in the atlanto-occipital assimilation group or the

RA group. These groups did show pseudotumor regression when measured visually and the statistical non-significance might be due to the small sample size.

Uniform surgical treatment plan had been made for huge number of patients with AAI despite various underlying symptom complexes. This could be owing to the surgeon’s immediate concern focused on AAI. Current consensus on treatment of choice for AAI patients is posterior fixation of the involved cervical segments.[16-19] Once AAI has occurred and there is evidence of myelopathy in MRI or there are signs of myelopathy, surgical decision should be made promptly in order to further prevent instability and neurological deficit. Based on this clinical thinking, our surgeon made customized surgical plans for each patient regarding fixation levels and detailed surgical technique. As a result all patients showed retro-odontoid pseudotumor regression and there was a favorable surgical outcome with statistical significance. Thus we could assume that there is a mechanical factor influencing retro-odontoid pseudotumor formation.

Yonezawa et al. described three types of retro-odontoid masses in RA patients regarding SI on T2W MRI: pannus type (low T1 and high T2 SI), pseudotumor type (low T1 and low T2 SI), and mixed type (low T1 and mixed T2 SI).[20] It is generally thought that the majority of patients with RA have a pannus-type retro-odontoid mass showing high T2 SI, which indicates the presence of an inflammatory component. Retro-odontoid pseudotumor with low T2 SI was thought to be more common in non-RA patients and its low T2 SI is based on the fibrous component.[21,22] Our study comprised retro-odontoid pseudotumor with all three aforementioned types (low T1 and low T2 SI, n = 24; low T1 and mixed T2 SI, n = 13; low T1 and high T2 SI, n = 1). The SI of retro-odontoid pseudotumor can be variable and nonspecific depending on the proportion of the tissue components contained. Our study showed that retro-odontoid pseudotumor regressed after posterior fixation irrespective of its T2 SI on MRI. Therefore, we suspect that immobilization of the atlantoaxial joint decreased mechanical stress, friction, and inflammatory process in structures such as the synovial capsule and transverse ligament.

One patient with AAI of unknown cause (case no. 12) had to undergo re-operation with a trans-oral approach because of insufficient regression of the retro-odontoid pseudotumor after posterior fixation alone. This patient had a pseudotumor of exceptionally large size with severe compressive myelopathy at initial presentation and the surgeon had planned a staged operation in advance. The initial size of the pseudotumor exceeded the sagittal diameter of the dens. Even though the sagittal diameter of the pseudotumor had decreased on postoperative MRI after 3 months, the patient underwent a second operation for removal of the retro-odontoid pseudotumor. The pathology report of the resected pseudotumor showed degenerative cartilage and granulation tissue.

There are several limitations to this study. First, this is retrospective research based on clinical data and imaging findings from EMRs and PACS. Even though we underwent a thorough review of medical records, some portion of the patients may have been misclassified as AAI of unknown cause. Furthermore, six out of 38 patients with retro-odontoid pseudotumor were lack of JOA scores. However, all 38 patients had NDI percentage and 32 patients had JOA scores. Thus, we believe that lack of a few patients' JOA scores could not have influenced our study result. Second, there were not enough samples allocated to each disease group and this could have affected some of non-significant statistical results. Third, pre- and postoperative image comparisons were performed using both CT and MRI. This may have resulted in measurement differences among image modalities. However, there was no significant difference in the measured size of the pseudotumor between CT and MRI when compared in the same patient. Therefore, we doubt that this may have influenced our study's results. CT and MRI can both be sufficient image modalities to observe the regression of pseudotumor after posterior fixation.

In conclusion, our study has shown that the incidence proportion of retro-odontoid pseudotumor in patients with symptomatic AAI was 23.2% (adult patients, 24%; pediatric patients, 16.7%). All patients who underwent posterior fixation showed regression of the pseudotumor despite various underlying diseases and SIs of the pseudotumors on initial MRI. We conclude that posterior fixation can be an appropriate surgical approach for patients with retro-odontoid pseudotumor and AAI irrespective of various underlying diseases.

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Tables

Table 1. Demographics of the Final Study Group and the Incidence of Retro−odontoid Pseudotumor in Patients with Atlantoaxial Instability

	Pseudotumor (+)	Pseudotumor (−)	Total
Male:Female	13:25	47:79	60:104
Mean age (± SD)	57 (± 18)	44 (± 17)	47 (± 18)
Age group			
Adult (> 18 years)	35 (24)	111 (76)	146
Pediatric (0–18 years)	3 (16.7)	15 (83.3)	18
Sx complex or Dx in adult patients			
AAI of unknown cause	11 (30.6)	25 (69.4)	36
Os odontoideum	11 (20.8)	42 (79.2)	53
Dens fracture	6 (26.1)	17 (73.9)	23
A−O assimilation	3 (42.9)	4 (57.1)	7
RA	3 (21.4)	11 (78.6)	14
Miscellaneous*	1 (7.7)	12 (92.3)	13

Sx complex or Dx in pediatric patients			
AAI of unknown cause	1 (50)	1 (50)	2
Os odontoideum	1 (12.5)	7 (87.5)	8
Dens fracture	0 (0)	4 (100)	4
A–O assimilation	1 (100)	0 (0)	1
RA	0 (0)	0 (0)	0
Miscellaneous†	0 (0)	3 (100)	3
Total	38 (23.2)	126 (76.8)	164

Values in parenthesis indicate percentage, except for age; SD indicates standard deviation; Sx indicates symptom; Dx indicates diagnosis; AAI indicates atlantoaxial instability; A–O assimilation indicates atlanto–occipital assimilation; RA indicates rheumatoid arthritis; Miscellaneous* include Morquio syndrome (n=1), multiple C1–2 fractures other than dens fracture (n = 5), congenital block vertebra C1–2 (n = 1), basiooccipital hypoplasia (n = 1), pseudogout (n = 1), psoriatic arthritis (n = 1), diffuse idiopathic skeletal hyperostosis (DISH) (n = 1), Gorham's disease (n = 1), and asymmetrical collapse of the C1–2 facet joint (n = 1); Miscellaneous† indicates atlanto–axial rotatory subluxation (n = 3).

Table 2. Clinical and Radiographic Characteristics of Patients with Retro-odontoid Pseudotumor

					Pseudotumor measurement *		Pseudotumor regression †		
Case No.	Age	Sex	Sx complex or Dx	CCS degree	Pre (mm)	Post (mm)	Regression (%)	Fusion level	F/U (mo)
1	39	M	AAI of unknown cause	1	14.9	13.4	53.4	C1-2	102
2	69	F	AAI of unknown cause	2	15.7	13.6	55.9	C1-2	5
3	64	F	AAI of unknown cause	1	14.1	13.2	33.3	C1-2	6
4	4	F	AAI of unknown cause	1	14.4	9.9	83.9	C1-2	19
5	67	M	AAI of unknown cause	2	14.6	13.3	58.5	C1-2	17
6	62	F	AAI of unknown cause	1	15.9	14.6	100	C1-2	12
7	68	F	AAI of unknown cause	3	17.4	12.1	72.4	C1-2	40
8	64	F	AAI of unknown cause	1	12.8	12.4	54.8	C1-2	2
9	68	M	AAI of unknown	1	20.1	16	59.7	C1-2	13

			cause						
10	66	F	AAI of unknown cause	3	21.2	16.6	51.1	C1-2	6
11	68	F	AAI of unknown cause	3	14.8	13.7	65.9	C1-2	8
12	72	F	AAI of unknown cause	3	24.9	23.5	17	C1-2	3
13	13	M	Os odontoideum	3	21.4	18.8	100	C1-2	29
14	69	F	Os odontoideum	3	22.3	17.8	50.6	C1-2	28
15	71	F	Os odontoideum	3	23.3	16.1	100	O-C3	15
16	54	M	Os odontoideum	3	19.8	17.5	100	C1-2	12
17	65	F	Os odontoideum	3	20.5	17.4	100	C1-2	2
18	60	M	Os odontoideum	3	14.5	11.6	58	C1-2	2
19	48	F	Os odontoideum	3	19.2	16.8	42.9	C1-2	2
20	31	M	Os odontoideum	3	18.8	16.8	100	C1-2	31
21	62	F	Os odontoideum	3	20.6	17.9	100	C1-2	1
22	59	M	Os odontoideum	3	19.5	17.3	49.2	C1-2	1
23	56	F	Os odontoideum	3	12.9	12.1	53.7	C1-2	1
24	71	M	Os odontoideum	3	25.2	19.2	64.7	C1-2	12
25	62	M	Dens fracture	3	17.5	15.4	61.9	C1−2	12
26	63	M	Dens fracture	3	23.8	20.8	60	C1-2	6
27	60	F	Dens fracture	3	17.5	14	100	C1-2	13
28	60	M	Dens fracture	3	15.9	12.6	100	C1-2	12

29	71	M	Dens fracture	3	18.7	14.4	100	C1-2	1
30	54	F	Dens fracture	3	12.3	11.7	69.8	C1-2	1
31	56	F	A-O assimilation	3	19.8	15.5	100	O-C2-C4-C5	8
32	8	F	A-O assimilation	3	11.5	8.9	100	O-C2	8
33	54	F	A-O assimilation	3	17.8	13.8	100	O-C2	1
34	70	F	A-O assimilation	3	16.9	14.8	100	O-C2	1
35	71	F	RA	2	15.2	13.6	50	C1-2	13
36	60	F	RA	2	16.7	14.1	53.8	C1-2	12
37	66	F	RA	3	16.6	14.2	59.5	C1-2	2
38	24	F	Morquio syndrome	3	14.7	12.2	58.9	C1-2	16

Sx, symptom; Dx, diagnosis; CCS indicates cervical central canal stenosis; Pre, preoperative; Post, postoperative; AAI indicates atlantoaxial instability; A-O assimilation indicates atlanto-occipital assimilation; Pseudotumor measurement* indicates largest sagittal diameter, from the anterior border of the odontoid process to the posterior border of the pseudotumor; Pseudotumor regression† indicates regression of the posterior portion of the pseudotumor, measured from the posterior border of the odontoid process to the posterior border of the pseudotumor; F/U, follow-up period; mo, months.

Table 3. Regression of the Retro-odontoid Pseudotumor in Each Symptom Complex or Diagnosis after Posterior Fixation

		Pseudotumor measurement*			Pseudotumor regression†
Sx complex or Dx	n	Pre (mm)	Post (mm)	*P* value	Regression (%)
AAI of unknown cause	12	16.7	14.4	0.002	58.8
Os odontoideum	12	19.8	16.6	0.002	76.6
Dens fracture	6	17.6	14.8	0.028	82
A-O assimilation	4	16.5	13.3	0.068	100
RA	3	16.2	14	0.1	54.5
Morquio syndrome	1	14.7	12.2	NA	58.9
Total	38	17.7	14.9	< 0.001	72.1

Sx, symptom; Dx, diagnosis; Pre, preoperative; Post, postoperative; AAI indicates atlantoaxial instability; A-O assimilation indicates atlanto-occipital assimilation; RA indicates rheumatoid arthritis; Pseudotumor measurement* indicates largest sagittal diameter, from the anterior border of the odontoid process to the posterior border of the pseudotumor; Pseudotumor regression † indicates regression of the posterior portion of the pseudotumor, measured from the posterior border of the odontoid process to the posterior border of the pseudotumor; NA indicates not applicable.

Table 4. Postoperative Improvement of Disease Severity Scores (JOA and NDI) in Patients with Retro-odontoid Pseudotumor

	Preoperative	Postoperative (6mo)	*P* value
Average JOA score (mean ± SD)	11.3 ± 3.2	13.4 ± 2.9	0.007
Average NDI score (mean ± SD)	41.5 ± 18.1	28.3 ± 15.2	< 0.001

JOA, Japanese Orthopedic Association; NDI, neck disability index; SD, standard deviation; mo, months.

Table 5. Postoperative Recovery Rate of Japanese Orthopedic Association (JOA) Scores in Patients with Retro-odontoid Pseudotumor

Recovery rate*	Number of patients
Excellent group	9 (28.1)
Good group	4 (12.5)
Fair group	5 (15.6)
Unchanged group	8 (25.0)
Worse group	6 (18.8)
Total	32 (100)

Recovery rate* was calculated using the following equation: (postoperative score − preoperative score)/(17 − preoperative score) x100; The recovery rate were classified into five groups: excellent (75 % < to ≤ 100 %), good (50 % < to ≤ 75 %), fair (25 % < to ≤ 50 %), unchanged (0 % < to ≤ 25 %), and worse (< 0 %); Values in parenthesis indicate percentage.

Figures

Figure 1. Examples of pseudotumor size measurement on MRI (A) and CT (B) respectively.

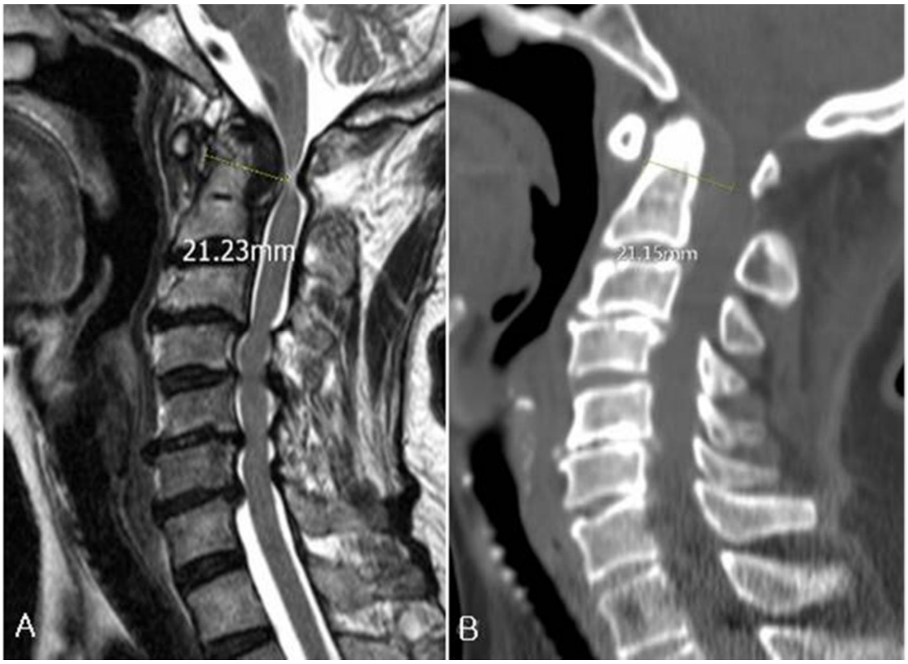


Figure 2. A 68-year-old female with atlantoaxial instability of unknown cause. There is posteriorly bulging retro-odontoid pseudotumor with mixed SI in the preoperative sagittal T2WI, causing compressive myelopathy (A). Pseudotumor nearly disappeared 40 months after C1-2 fixation (B).

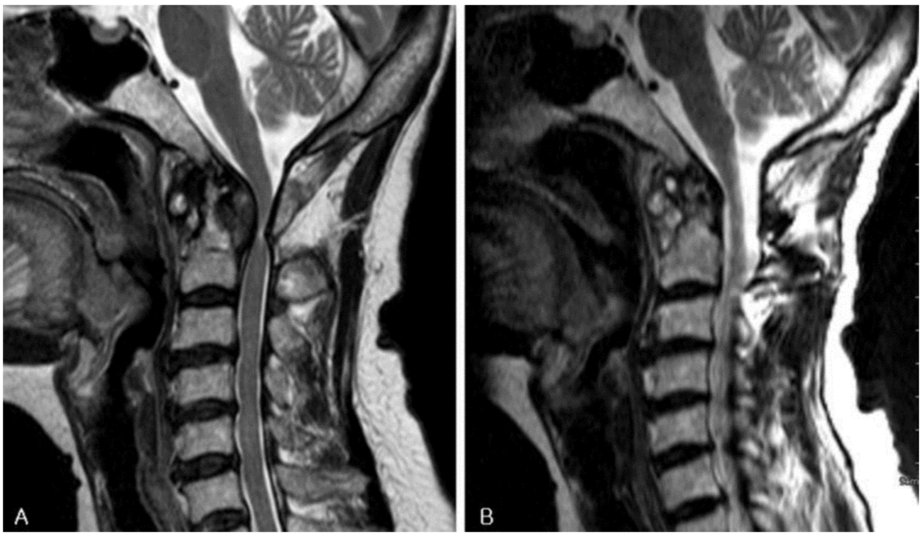


Figure 3. A 71-year-old female with os odontoideum. Retro-odontoid pseudotumor with T2 low SI is causing compressive myelopathy in the preoperative MRI (A). 15 months after occipito-cervical fixation, pseudotumor has nearly disappeared (B).

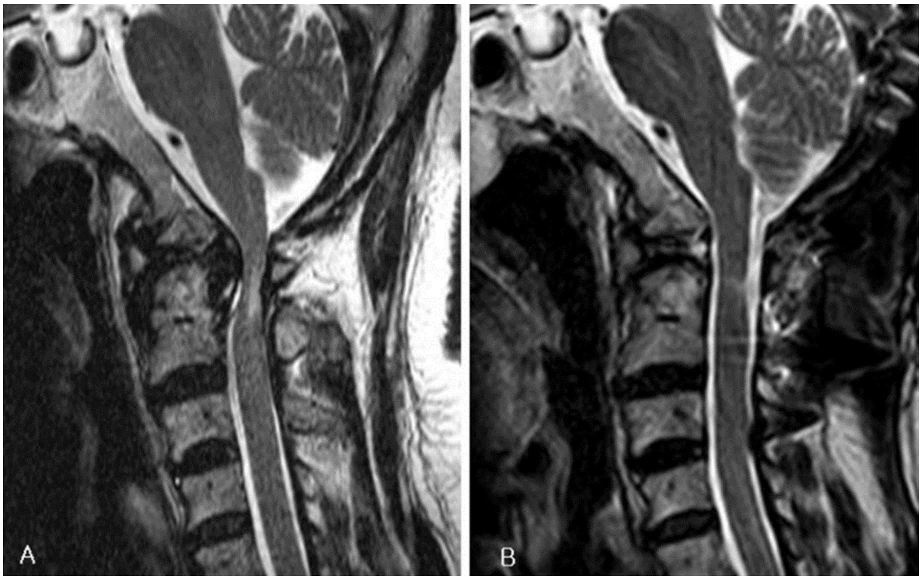
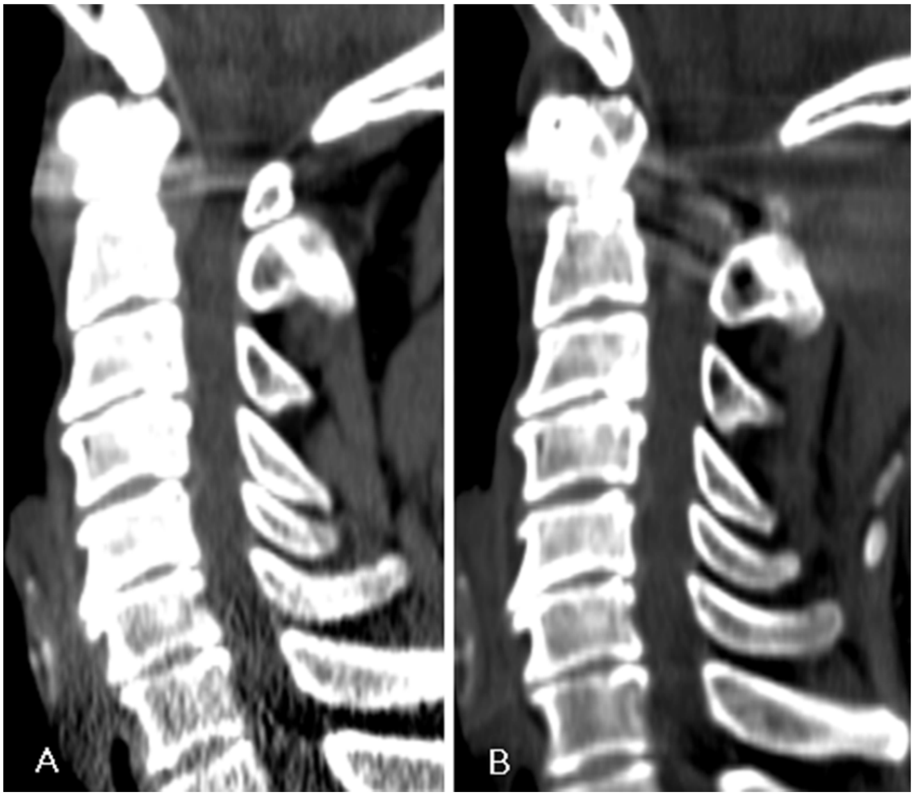


Figure 4. A 63−year−old male with old dens fracture. Retro−odontoid high attenuated mass representing pseudotumor is well−delineated in the preoperative CT (A). 6 months after C1-2 fixation, pseudotumor regression is noted in postoperative CT (B).



논 문 초 록

목적: 환추축 불안정성이 있는 환자들에서 치상돌기 후방부의 가성 종양의 발생률을 알아보고자 하며 가성 종양이 있는 환자들에서 후방 고정 수술 후 가성 종양의 크기가 감소하는지 확인하고자 한다.

대상 및 방법: 2004 년부터 2015 년까지 환추축 불안정성으로 후방 고정 수술을 받은 환자들 중 1-2 번 경추에 이전 수술 병력이 있거나 종양 혹은 감염성 질환의 기록이 없는 환자들을 연구 집단에 포함시켰다. 연구 집단에 포함된 환자들은 기저질환과 (rheumatoid arthritis, os odontoideum, atlanto-occipital assimilation, dens fracture, atlantoaxial instability of unknown cause, etc.) 나이에 (adult and pediatric groups) 따라 분류하였고 각 집단에서 치상돌기 후방부의 가성 종양 발생률을 확인하였다. 가성 종양이 발견된 환자들에게서 수술 전 후 자기 공명 혹은 컴퓨터 단층 촬영 영상을 통해 가성 종양의 크기를 비교하였으며 가성종양의 크기는 치상돌기의 전면부터 가성 종양의 후면까지를 측정한 값으로 정하였다.

결과: 164 명 중 38 명의 환자들에서 치상돌기 후방부 가성 종양이 확인되었다 (23.3%). 38 명 중 세 명은 rheumatoid arthritis 환자들이었고 나머지는 다양한 질환 군의 환자들이었다. 가성 종양의 크기는 38 명의 환자들에서 모두 통계적으로 유의미하게

감소하였다. 치상돌기 전면부터 가성 종양의 후면까지를 측정한 가성 종양의 크기는 수술 전 평균 17.7 mm 에서 수술 후 14.9 mm 로 감소하였다 ($P < 0.001$).

결론: 다양한 기저 질환을 가진 환추축 불안정성 환자들에게서 치상돌기 후방부의 가성 종양 발생률은 23.2%로 확인되었다. 치상돌기 후방부 가성 종양은 기저질환과 상관 없이 후방 고정 수술 후 크기 감소를 보였다.

주요어: 치상돌기 후방부 가성 종양, 환추축 불안정성, 후방 고정 수술, 자기 공명 영상, 컴퓨터 단층 촬영

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