C2 Radiculopathy Due to a Retro-Odontoid Pseudotumor: A Case Report

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A 69-year-old man with a nine-month history of crawling sensation and two-month history of pain in the left occipital region with a visual analog scale (VAS) of 79.5 mm presented at our institution. Neurological findings showed no muscle weakness in upper and lower extremities; however, biceps tendon reflex on the right and patellar reflexes in both limbs were increased. Dynamic radiography of the cervical spine revealed that atlantodental interval (ADI) was 2.2 mm in the flexed position and 1.2 mm in the extension one.

The range of motion on C1/2 was 12°, indicating that there was no instability in the C1/2 joint (Fig. 1A and B). Magnetic resonance imaging (MRI) revealed a retro-odontoid pseudotumor with iso-intensity on T1-weighted imaging (Fig. 2A), hypointensity on T2-weighted imaging (Fig. 2B), and rim enhancement with gadolinium (Fig. 2C). The pseudotumor extended toward the left ventral aspect and compressed the left C2 nerve root (Fig. 2D and E). Celecoxib and pregabalin were prescribed for the pain, and nerve root

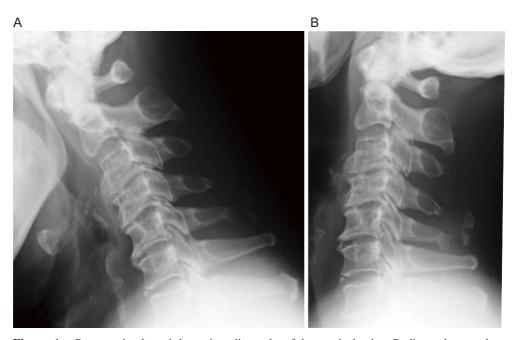


Figure 1. Preoperative lateral dynamic radiographs of the cervical spine. Radiographs reveal no joint instability at C1/2. (A) Flexed position. (B) Extension position.

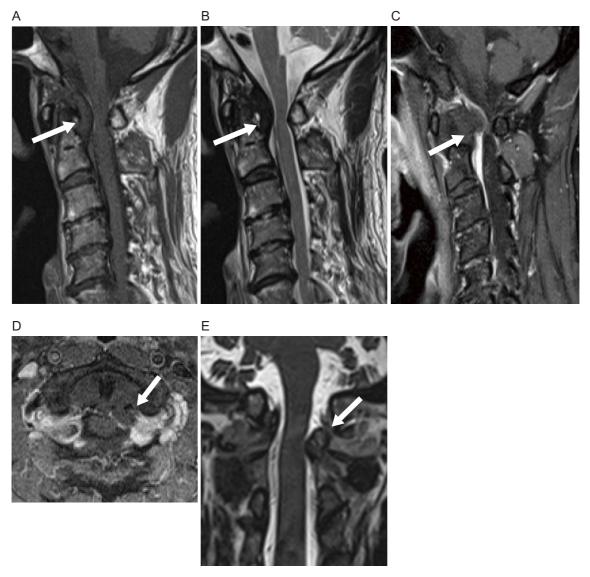


Figure 2. Preoperative magnetic resonance imaging. (A) Retro-odontoid pseudotumor is iso-intense on T1-weighted sagittal image (*arrow*), (B) Retro-odontoid pseudotumor is hypointense on T2-weighted sagittal image (*arrow*). (C) Retro-odontoid pseudotumor has rim enhancement on T1-weighted gadolinium-enhanced sagittal image (*arrow*). (D) T1-weighted gadolinium-enhanced axial image reveals the pseudotumor extending toward the left ventral aspect (*arrow*). (E) T2-weighted coronal image shows compression of the left nerve root of C2 by the pseudotumor (*arrow*).

blocks were severally performed at C2 under echographic guidance. Although he experienced temporary relief after each nerve root block, the symptoms consistently recurred; therefore, the patient opted for surgery.

Laminectomy of the posterior arch of the atlas revealed that the pseudotumor extended between the bifurcation of the left C2 nerve root and spinal cord. Moreover, the left C2 nerve root was compressed by the pseudotumor (Fig. 3A and B). After partial resection of the pseudotumor, decompression of the left C2 nerve root was confirmed. Histopathologically, the pseudotumor included cartilaginous tissue. Four months postoperatively, the crawling sensation and pain in the left occipital region had significantly resolved, and the VAS score was 4 mm. Dynamic radiography revealed that ADI was 2.7 mm in the flexed position, indicat-

ing that there was no progression of C1/2 instability. Postoperative MRI showed that there was no re-growth of the pseudotumor; symptoms had not recurred at the last follow-up visit, 9 months after surgery.

The retro-odontoid pseudotumor was first reported by Sze et al.¹⁾ in 1986. It is established that rheumatoid arthritis and subluxation of the C1/2 joint that results in instability of the C1/2 joint often cause retro-odontoid pseudotumors. Retro-odontoid pseudotumors cause cervical myelopathy due to compression of the spinal cord. However, C2 radiculopathy due to retro-odontoid pseudotumor is extremely rare. Overall, the prevalence of C2 radiculopathy is reported to be 3.2 per 100,000²⁾. The etiology of C2 radiculopathy could be vascular (irritation by intra/extracranial vessels or arteritis), neurogenic (schwannomas, myelitis, or radiculoneuritis), or

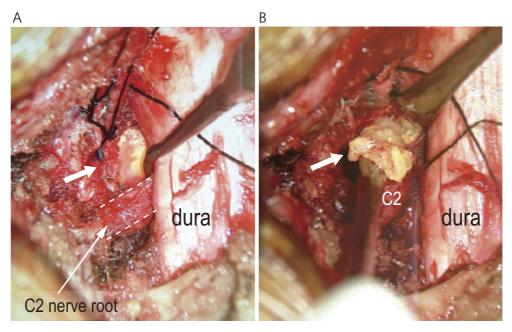


Figure 3. Intraoperative photographs. (A) Left nerve root of C2 (*break lines*) is compressed by the pseudotumor (*arrow*). (B) The pseudotumor (*arrow*) is resected partially.

bony (arthrosis or callus formation after C1/2 fractures)³⁾. A case of C2 radiculopathy due to retro-odontoid pseudotumor was reported by Taniguchi et al.⁴⁾ in the Japanese literature. To the best of our knowledge, there have been no reports on C2 radiculopathy due to retro-odontoid pseudotumors in the English literature.

Spinal fusion surgery is a surgical intervention for retroodontoid pseudotumors, and favorable clinical outcomes can be achieved⁵⁻⁷⁾. Taniguchi et al.⁴⁾ performed laminectomy of the atlas and spinal fusion for retro-odontoid pseudotumors. Conversely, laminectomy without fusion could further yield favorable clinical outcomes in patients with retro-odontoid pseudotumors (who have less C1/2 joint instability)^{6,8-10)}. In the present case, C1/2 joint instability was not noted; therefore, partial resection of the pseudotumor by laminectomy of the atlas without spinal fusion was performed.

Although no progress in the instability of the C1/2 joint or pseudotumor re-growth was observed 9 months after surgery, this patient requires long-term follow-up. In conclusion, partial resection of the pseudotumor and decompression of the C2 nerve root without spinal fusion were effective in C2 radiculopathy due to a retro-odontoid pseudotumor.

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Ethical Approval: Not applicable.

Informed Consent: Written informed consent was obtained from the participant in this case report.

References

- **1.** Sze G, Brant-Zawadzki MN, Wilson CR, et al. Pseudotumor of the craniovertebral junction associated with chronic subluxation: MR imaging studies. Radiology. 1986;161(2):391-4.
- **2.** Koopman JS, Dieleman JP, Huygen FJ, et al. Incidence of facial pain in the general population. Pain. 2009;147(1-3):122-7.
- Choi I, Jeon SR. Neuralgias of the head: occipital neuralgia. J Korean Med Sci. 2016;31(4):479-88.
- Taniguchi S, Takahashi H, Terashima F, et al. [A case report of radiculopathy due to retro-odontoid pseudotumor]. J East Jpn Orthop Traumatol. 2014;26(3):395. Japanese.
- **5.** Chikuda H, Seichi A, Takeshita K, et al. Radiographic analysis of the cervical spine in patients with retro-odontoid pseudotumors. Spine. 2009;34(3):E110-4.
- **6.** Kobayashi K, Imagama S, Ando K, et al. Post-operative regression of retro-odontoid pseudotumors treated with and without fusion. Eur Spine J. 2018;27(12):3105-12.
- Sono T, Onishi E, Matsushita M. Radiographic risk factors and surgical outcomes for retroodontoid pseudotumors. J Spinal Disord Tech. 2014;27(6):E193-8.
- Kakutani K, Doita M, Yoshikawa M, et al. C1 laminectomy for retro-odontoid pseudotumor without atlantoaxial subluxation: review of seven consecutive cases. Eur Spine J. 2013;22(5):1119-26.

- **9.** Shimizu T, Otsuki B, Fujibayashi S, et al. Incidence and risk factors of anterior arch fracture of the atlas following C1 laminectomy without fusion. Spine. 2018;43(10):667-74.
- 10. Takemoto M, Neo M, Fujibayashi S, et al. Clinical and radio-graphic outcomes of C1 laminectomy without fusion in patients with cervical myelopathy that is associated with a retro-odontoid pseudotumor. Clin Spine Surg. 2016;29(10):E514-21.

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