

Retro-odontoid cystic mass treated by laminectomy and C1-C2 fixation

Dasheng Lin, Zhenqi Ding, Yanjie Guo, Kejian Lian

ABSTRACT

Retro-odontoid cysts associated with chronic atlantoaxial subluxation are extremely rare. This article describes a case of retro-odontoid cystic mass associated with chronic atlantoaxial subluxation and its management with posterior C1 and partial C2 laminectomy and C1-C2 pedicle screw fixation without resection of the retro-odontoid cyst. A 64-year-old woman experienced a sudden onset of neck pain, hand and foot paresthesia. Atlantoaxial instability associated with a retro-odontoid cystic mass was found in the imaging. The patient underwent posterior C1 and partial C2 laminectomy and C1-C2 pedicle screws fixation without resection of the retro-odontoid cyst. During the 24 months followup period, the cyst disappeared completely and the patient remained symptom free and returned to independent daily living. These findings suggest that posterior laminectomy and fixation without resection of the retro-odontoid cyst is relatively simple and safe and the results are satisfactory.

Key words: Atlantoaxial subluxation, atlantoaxial instability, retro-odontoid cyst, posterior fixation

MeSH terms: Spine, cervical vertebrae, spinal diseases, orthopedic equipment, joint instability

INTRODUCTION

Retro-odontoid cystic mass associated with chronic atlantoaxial subluxation is extremely rare. They are represented in the literature predominantly as case reports. The etiology of the cysts remains controversial. Takeuchi *et al.*¹ explained that the transverse ligament becomes degenerated and hypertrophic because of chronic mechanical stress by atlantoaxial subluxation. Then, a part of the ligament develops reactive granulation tissue with a small vessel formation. Finally, rupture of these small vessels cause repeated episodes of microbleeding, resulting in formation of a cyst. Direct excision of the cysts and fixation has commonly been performed for the surgical treatment of retro-odontoid cystic mass associated with chronic atlantoaxial subluxation.¹⁻⁵ However, when the cyst is located in the ventral side of

spinal cord, direct resection is technically dangerous. Here, we present a case of a retro-odontoid cyst associated with chronic atlantoaxial subluxation and located in the ventral side of spinal cord. The cystic mass disappeared after posterior C1 and partial C2 laminectomy and C1-C2 pedicle screws fixation without resection of the retro-odontoid cyst.

CASE REPORT

A 64 year old woman reported experiencing a sudden onset of neck pain, hand and foot paresthesia when she woke early in the morning. She was sent to a local hospital, where she was treated conservatively with external stabilization of the neck and bed rest. At 2 weeks later, the patient was transferred to our hospital because her symptoms had not relieved. There were no clinical or laboratory findings of rheumatoid arthritis. There was no history of head and neck trauma. X-ray showed that the atlantoaxial instability [Figure 1a-c]. Computed tomography revealed an abnormality of the odontoid process [Figure 1d and e]. Magnetic resonance imaging detected an oval retro-odontoid cystic mass, which compressed the spinal cord. The mass showed uniform low intensity on T1-weighted image [Figure 1f] and uniform high signal intensity on T2-weighted image [Figure 1g].

The surgery was carried out with the patient positioned prone with the head slightly flexed and a midline incision was made. Subsequently, paraspinal muscles were peeled to expose the bilateral laminae of C1 and C2. Magerl's method was used to place pedicle screws on C1 and C2.

Department of Orthopaedic Surgery, The Affiliated Southeast Hospital of Xiamen University, Orthopaedic Center of People's Liberation Army, Zhangzhou 363000, China

Address for correspondence: Dr. Kejian Lian, Department of Orthopaedic Surgery, The Affiliated Southeast Hospital of Xiamen University, Orthopaedic Center of People's Liberation Army, Zhangzhou 363000, China. E-mail: enschor98@126.com

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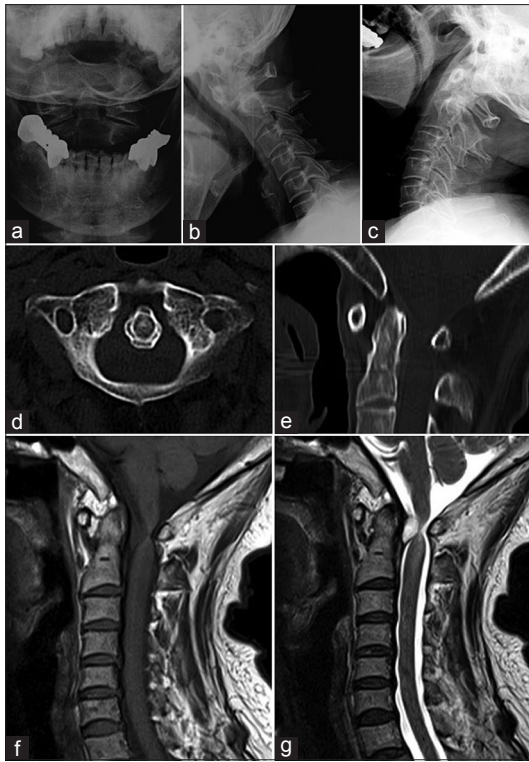


Figure 1: (a-c) Preoperative open-mouth anteroposterior radiograph and dynamic flexion and extension radiograph showing the atlantoaxial instability. (d and e) Preoperative computed tomography revealed an abnormality of the odontoid process. (f) The mass showed uniform low intensity on T1-weighted image. (g) The mass showed uniform high signal intensity on T2-weighted image

The laminectomy of C1 and partial C2 were performed, and then the neck was slightly extended to reduce the atlanto-axial joint. A pedicle screw system was used for fixation. Finally, an autogenous iliac crest graft was implanted in to C1 and C2 bilateral joints to facilitate fusion. Spinal cord monitoring was available during the procedures.

Postoperatively, neck pain immediately disappeared and neurologic symptoms disappeared by 1 week. The patient was ordered to wear a cervical collar for 3 months for protection. X-ray [Figure 2a and b], computed tomography [Figure 2c] and magnetic resonance imaging [Figure 2d] were taken on the day of surgery. At 24 months after surgery, the retro-odontoid cystic mass was no longer present [Figure 2e], and the patient remained symptom free and returned to independent daily living. Magnetic resonance imaging scans at 24 months after surgery showed that atlantodental interval was wider, compared to the day of surgery. There was complete bony union of C1-C2.

DISCUSSION

An atlantoaxial subluxation associated with a non tumorous lesion in the posterior region of the odontoid process had

been described as a retro-odontoid cyst or retro-odontoid pseudotumor in the initial report by Sze *et al.*⁶ It usually occurs in the elderly, without clear association to any antecedent trauma. The natural history is that of progressive cervical myelopathy. Most patients are between 60- and 80-years-old, with both sexes being equally affected. All of these patients are seronegative for rheumatoid arthritis or other inflammatory arthropathies.⁷ However, the etiology of the cysts remains controversial. In the postulated view on its pathomechanism, preexisting atlantoaxial instability was presumed to cause repeated tear and subsequent hypertrophy of the transverse ligament, thus leading to the formation of the pseudotumor.⁸ Takeuchi *et al.*¹ explained that the transverse ligament of axis became degenerated and hypertrophic because of chronic mechanical stress by atlantoaxial subluxation. Then, a part of the ligament developed reactive granulation tissue with small vessel formation. Finally, rupture of these small vessels caused repeated episodes of microbleeding, resulting in formation of a cyst.

Optimal treatment for a retro-odontoid cystic mass associated with chronic atlantoaxial subluxation has not been established. Direct excision of the cysts have commonly been performed for the surgical treatment. The surgical approach may be anterior or posterior. The cysts are always located in the midline directly in front of the spinal cord. Transoral or anterolateral decompression is theoretically the optimal option.²⁻⁵ Takeuchi *et al.*¹ reported a case that they incised the dura to reach the mass and resect through the posterior approach. However, the anterior and posterior surgical excision involve a lot of complex technical problems and requires more operating time and blood losses, higher risks and complications and should be performed only by an experienced surgical team. It is also reported that posterior fixation of such degenerative tissue would reduce repetitive mechanical stimulation, thereby inhibiting stress on the spinal cord and reducing the inflammation which had produced the degenerative lesions.^{9,10} Similar to degenerative cysts, retro-odontoid hypertrophy of soft tissue mass in a non rheumatoid patient with atlantoaxial subluxation has been reported to decrease after posterior fixation.¹¹⁻¹⁵ Nevertheless, the surgical procedure is appropriate for no neurologic symptoms and the small cystic mass. For patients with large cystic mass leading to severe canal stenosis, the available space required for spinal cord is very limited. During the process, the atlanto-axial joint reduction is prone to trauma to the spinal cord, which might cause spinal damage or even paraplegia.

We selected posterior C1 and partial C2 laminectomy and C1-C2 pedicle screw fixation without resection of the retro-odontoid cyst in the patient because the cyst

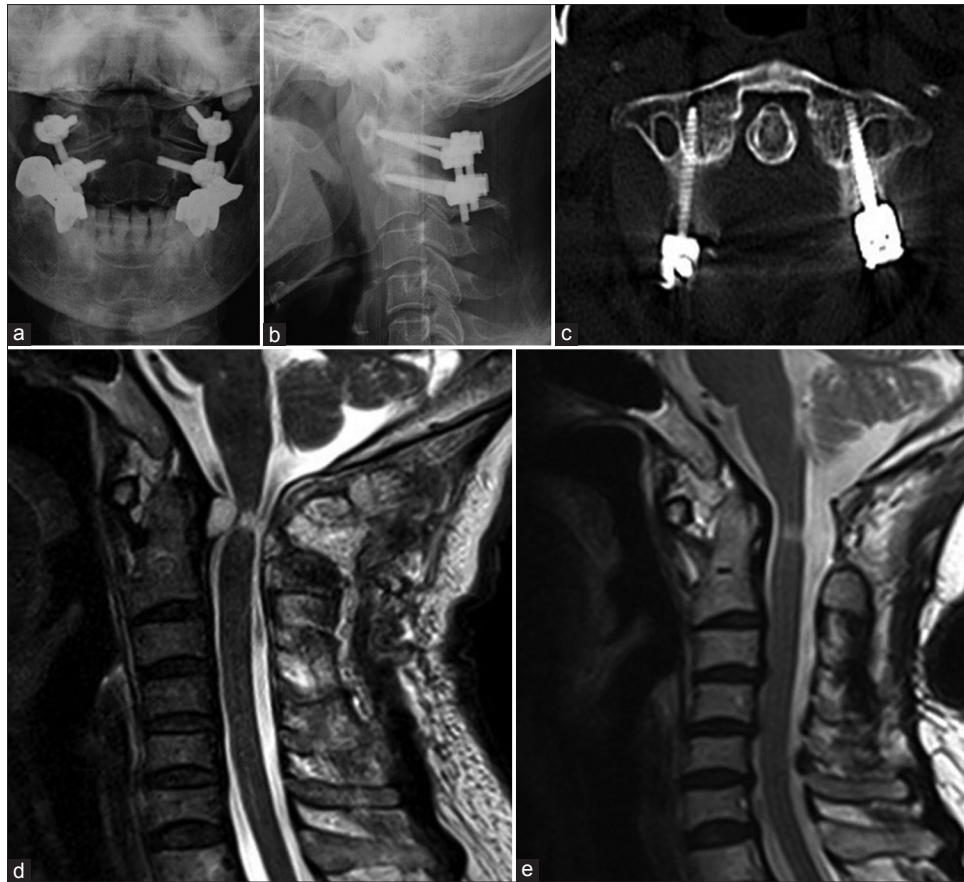


Figure 2: (a and b) Postoperative open mouth anteroposterior radiograph and lateral radiograph showing the rigid fixation between C1-C2 and atlantoaxial joint had been reset. (c) Postoperative computed tomography showed that atlantoaxial joint had been reset. (d) Postoperative sagittal T2-weighted magnetic resonance image showed that the mass still compressed the spinal cord. (e) Sagittal T2-weighted magnetic resonance image taken 24 months after surgery showed that the mass was no longer present

was located in the ventral side of spinal cord and the neurological deficit was less obvious. The posterior arch of the atlas and partial laminae of C2 were resected, providing the available space for the spinal cord drift. As far as possible, avoid damaging the nervous system. C1-C2 pedicle screws fixation might provide immediate and long term stability of the cervical spine, and is helpful in preventing an increase in the size and also to shrink the cyst. During the 24 months followup period, the cyst disappeared completely, and the patient remained symptom free and returned to independent daily living. These findings suggest that posterior laminectomy and fixation without resection of the retro-odontoid cyst is relatively simple and safe, and the results are satisfactory when the large cyst was located in the ventral side of spinal cord and the neurological deficit was less obvious. However, if the neurological deficit is progressive and severe, then resection of the retro-odontoid cysts should be selected because it could take time for the pseudotumor to disappear.

REFERENCES

1. Takeuchi M, Yasuda M, Takahashi E, Funai M, Joko M, Takayasu M. A large retro-odontoid cystic mass caused by transverse ligament degeneration with atlantoaxial subluxation leading to granuloma formation and chronic recurrent microbleeding case report. *Spine J* 2011;11:1152-6.
2. Marbacher S, Lukes A, Vajtai I, Ozdoba C. Surgical approach for synovial cyst of the atlantoaxial joint: A case report and review of the literature. *Spine (Phila Pa 1976)* 2009;34:E528-33.
3. Choe W, Walot I, Schlesinger C, Chambi I, Lin F. Synovial cyst of dens causing spinal cord compression. Case report. *Paraplegia* 1993;31:803-7.
4. Birch BD, Khandji AG, McCormick PC. Atlantoaxial degenerative articular cysts. *J Neurosurg* 1996;85:810-6.
5. Tobenas-Dujardin AC, Derrey S, Proust F, Toussaint P, Laquerriere A, Freger P. Atlantoaxial synovial cyst. A case report. *Neurochirurgie* 2004;50:652-6.
6. Sze G, Brant-Zawadzki MN, Wilson CR, Norman D, Newton TH. Pseudotumor of the craniovertebral junction associated with chronic subluxation: MR imaging studies. *Radiology* 1986;161:391-4.
7. Cai CY, Palmer CA, Paramore CG. Exuberant transverse ligament degeneration causing high cervical myelopathy. *J Spinal Disord* 2001;14:84-8.
8. Jun BY, Yoon KJ, Crockard A. Retro-odontoid pseudotumor in diffuse idiopathic skeletal hyperostosis. *Spine (Phila Pa 1976)* 2002;27:E266-70.
9. Yoshida G, Kamiya M, Yukawa Y, Kanemura T, Imagama S,

- Matsuyama Y, *et al.* Rheumatoid vertical and subaxial subluxation can be prevented by atlantoaxial posterior screw fixation. *Eur Spine J* 2012;21:2498-505.
10. Nagaria J, Kelleher MO, McEvoy L, Edwards R, Kamel MH, Bolger C. C1-C2 transarticular screw fixation for atlantoaxial instability due to rheumatoid arthritis: A seven-year analysis of outcome. *Spine (Phila Pa 1976)* 2009;34:2880-5.
 11. Sagiuchi T, Shimizu S, Tanaka R, Tachibana S, Fujii K. Regression of an atlantoaxial degenerative articular cyst associated with subluxation during conservative treatment. Case report and review of the literature. *J Neurosurg Spine* 2006;5:161-4.
 12. Cihanek M, Fuentès S, Metellus P, Pech-Gourg G, Dufour H, Grisoli F. Disappearance of retro-odontoid pseudotumor after C1-C2 transarticular fixation screw. *Neurochirurgie* 2008;54:32-6.
 13. Watanabe M, Iwashina T, Sakai D, Yamamoto Y, Mochida J. Cervical myelopathy with retroodontoid pseudotumor caused by atlantoaxial rotatory fixation and senile tremor. *Tokai J Exp Clin Med* 2009;34:39-41.
 14. Ogata T, Kawatani Y, Morino T, Yamamoto H. Resolution of intraspinal retro-odontoid cyst associated with os odontoideum after posterior fixation. *J Spinal Disord Tech* 2009;22:58-61.
 15. Chikuda H, Seichi A, Takeshita K, Shoda N, Ono T, Matsudaira K, *et al.* Radiographic analysis of the cervical spine in patients with retro-odontoid pseudotumors. *Spine (Phila Pa 1976)* 2009;34:E110-4.

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