

CASE REPORT

Immediate Reduction of a Retro-odontoid Synovial Cyst Following Lateral Atlantoaxial Joint Puncture and Arthrography

A Case Report

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Study Design. Case report.

Objective. We report on a case with a retro-odontoid synovial cyst, and the immediate reduction of the cyst was confirmed after lateral atlantoaxial joint puncture and arthrography.

Summary of Background Data. Retro-odontoid synovial cysts are rare diseases located posteriorly to a dense axis. Because most reports have focused on surgical treatment, only a few have examined nonsurgical treatment. However, several months are required after nonsurgical treatment until cyst regression.

Methods. A 52-year-old female presented with atlantoaxial instability. She complained of neck pain and numbness in her hands. Magnetic resonance imaging revealed a retro-odontoid synovial cyst. Lateral atlantoaxial joint puncture and arthrography were performed.

Results. Two days after treatment, the patient showed significant improvement in the numbness of her hands, and a follow-up magnetic resonance imaging revealed an immediate reduction in the cyst. During a 4.5-year follow-up period, no recurrence of the clinical symptoms occurred.

Conclusion. Lateral atlantoaxial joint puncture may immediately reduce retro-odontoid synovial cysts, and the lateral atlantoaxial joint has a communication channel with the retro-odontoid synovial cyst via the atlantodental joint. Once disappearance of the cyst is confirmed, an acceptable long-term outcome can be achieved with nonsurgical treatment even in cases with atlantoaxial instability.

Key words: retro-odontoid synovial cyst, cervical spine, atlantoaxial joint, atlantodental joint, atlantoaxial joint puncture, atlantoaxial joint arthrography, nonsurgical treatment, atlantoaxial instability, spinal cord, communication channel.

Level of Evidence: N/A

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Retro-odontoid synovial cysts are rare diseases usually located posteriorly to a dense axis.^{1–4} To date, most reports have focused on surgical treatment,^{2–10} and only a few reports have examined nonsurgical treatment. However, several months are required after nonsurgical treatment until cyst regression.^{11,12} Herein, we report on a case with a retro-odontoid synovial cyst in which its immediate reduction was confirmed after lateral atlantoaxial joint puncture and arthrography. This case suggests that lateral atlantoaxial joint puncture may provide acceptable short- and long-term outcomes even in cases with atlantoaxial instability.

CASE REPORT

A 52-year-old female complained of neck pain since February 2009, and she began to notice numbness in her hands by November 2009. She presented in August 2010 complaining of increasing numbness in her hands. The neurological examination revealed hypesthesia to touch and pain in her hands with full motor strength. Magnetic resonance imaging demonstrated a large cystic mass located posteriorly to the odontoid process of C2 (Figure 1A, B). Flexion-extension myelography of the cervical spine demonstrated atlantoaxial instability and spinal cord compression at the C1–C2 levels. Computed tomographic myelography demonstrated a mass around the odontoid process. Thus, the patient was diagnosed with a retro-odontoid cyst and atlantoaxial instability.

In May 2010, we performed lateral atlantoaxial joint puncture and arthrography. An attempted puncture resulted in no fluid return. Arthrography demonstrated a communication channel between both sides of the lateral atlantoaxial joints and the cyst via the atlantodental joint (Figure 2A, B). Unexpectedly, 2 days subsequent to the puncture, the patient

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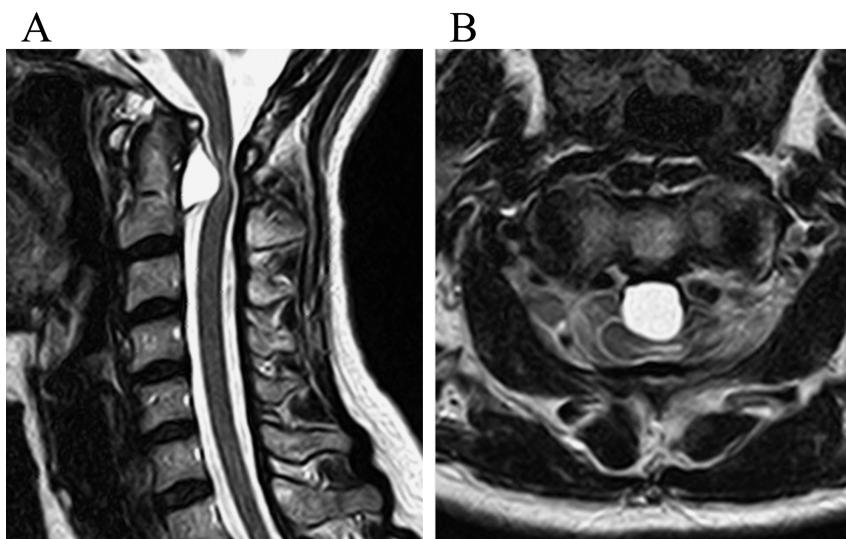


Figure 1. **A** and **B**, Sagittal and axial magnetic resonance images revealing a large cystic mass located posteriorly to the odontoid process of C2, which is severely compressing the spinal cord dorsally.

showed significant improvement in the numbness and hypoesthesia of her hands. A follow-up magnetic resonance imaging 1 month after treatment demonstrated a significant reduction in the retro-odontoid cyst (Figure 3A, B), and magnetic resonance imaging 5 months after puncture showed complete disappearance of the cyst (Figure 3C, D). During a 4.5-year follow-up period, disappearance of the cyst was confirmed, and no recurrence of the clinical symptoms and no deterioration in the atlantoaxial instability occurred.

DISCUSSION

This case demonstrated 2 clinical issues. First, lateral atlantoaxial joint puncture may immediately reduce retro-odontoid synovial cysts. Most reports on retro-odontoid synovial cysts focus on surgical treatment.²⁻¹⁰ Only 2 reports have demonstrated that conservative treatment with an external neck brace results in the regression of retro-odontoid cysts.^{11,12} However, clinical improvement after treatment with an external neck brace requires several months. To the best of our knowledge, this is the first reported case in which nonsurgical treatment (*i.e.*, lateral atlantoaxial joint puncture) immediately reduced the retro-odontoid synovial cyst.

Second, we demonstrated that the lateral atlantoaxial joint has a communication channel with the retro-odontoid synovial cysts *via* the atlantodental joint. Thus far, most reports on retro-odontoid synovial cysts have suggested that the cysts arise from atlantodental articulation and contact with the quadrate or transverse ligament of dens^{1,2,5,13} or the synovial joint between an os odontoideum and the body of C2.^{7,14} Few reports have demonstrated communication between the synovial cyst around the dense, lateral atlantoaxial joint and subarachnoid space.^{10,15} In our case, lateral atlantoaxial joint arthrography demonstrated communication between both sides of the lateral atlantoaxial joints and the cyst *via* the atlantodental joint. We speculate that the presence of this communication channel participates in the pathogenesis of retro-odontoid synovial cysts.

Little is known about the long-term outcomes of nonsurgical treatment of retro-odontoid synovial cysts. To date, the longest follow-up period for nonsurgical treatment of cysts was 2 years.¹¹ In our case, no recurrence of the clinical symptoms occurred after 4 years after disappearance of the cyst was confirmed. Therefore, it seems reasonable to think that once the disappearance of the cyst is confirmed, acceptable

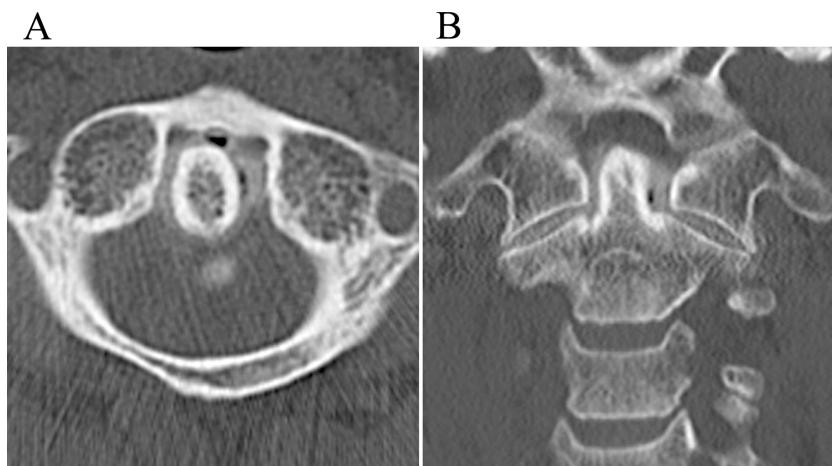


Figure 2. **A** and **B**, Computed tomograms after arthrography demonstrate a continuously spreading contrast medium into both sides of the lateral atlantoaxial and atlantodental joints and into the mass around the odontoid process.

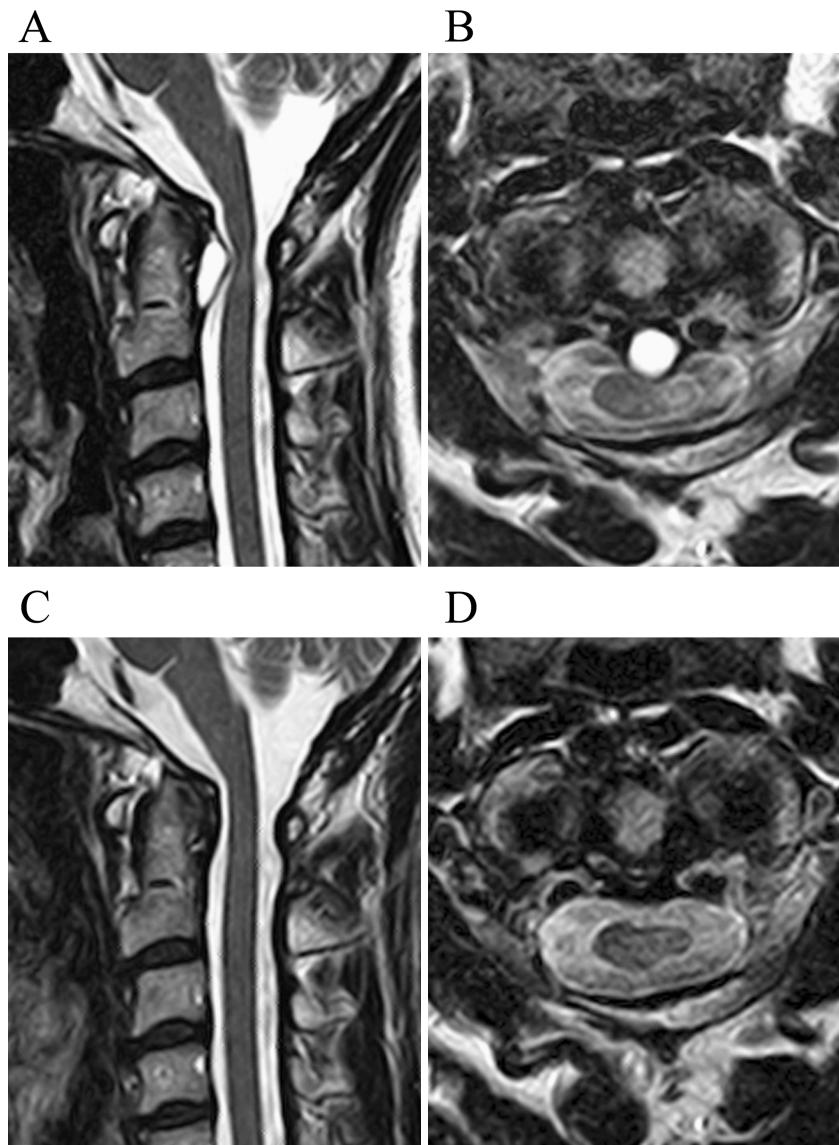


Figure 3. **A** and **B**, Sagittal and axial magnetic resonance images 2 months after the puncture demonstrating a significant reduction in the retro-odontoid cyst. **C** and **D**, Sagittal and axial magnetic resonance images 5 months after puncture showing complete disappearance of the retro-odontoid cyst.

long-term outcomes can be achieved with nonsurgical treatment even in cases with atlantoaxial instability.

In conclusion, lateral atlantoaxial joint puncture may immediately reduce retro-odontoid synovial cysts, and the lateral atlantoaxial joint has a communication channel with the retro-odontoid synovial cysts *via* the atlantodental joint. Thus, lateral atlantoaxial joint puncture may be a cost-effective and low-invasive alternative treatment of retro-odontoid synovial cysts in selected patients. Once disappearance of the cyst is confirmed, nonsurgical treatment can provide acceptable long-term outcomes even in cases with atlantoaxial instability. We think that lateral atlantoaxial joint puncture is worth performing before making the final decision to perform surgery. The presence of a communication channel between both sides of the atlantoaxial joint and the atlantodental joint may participate in the pathogenesis of retro-odontoid cysts. Further studies should be conducted to determine the pathogenesis and appropriate treatment strategy for retro-odontoid synovial cysts.

➤ Key Points

- Lateral atlantoaxial joint puncture may immediately reduce retro-odontoid synovial cysts.
- The lateral atlantoaxial joint has a communication channel with retro-odontoid synovial cysts *via* the atlantodental joint.
- Once the cyst's disappearance is confirmed, acceptable long-term outcomes can be achieved with nonsurgical treatment even in cases with atlantoaxial instability.

References

1. Onofrio BM, Mih AD. Synovial cysts of the spine. *Neurosurgery* 1988;22:642-7.
2. Choe W, Walot I, Schlesinger C, et al. Synovial cyst of dens causing spinal cord compression. Case report. *Paraplegia* 1993;31: 803-7.

3. Fransen P, Pizzolato GP, Otten P, et al. Synovial cyst and degeneration of the transverse ligament: an unusual cause of high cervical myelopathy. Case report. *J Neurosurg* 1997;86:1027–30.
4. Van Gompel JJ, Morris JM, Kasperbauer JL, et al. Cystic deterioration of the C1-2 articulation: clinical implications and treatment outcomes. *J Neurosurg Spine* 2011;14:437–43.
5. Birch BD, Khandji AG, McCormick PC. Atlantoaxial degenerative articular cysts. *J Neurosurg* 1996;85:810–6.
6. Akiyama H, Tamaki N, Kondoh T, et al. Craniocervical junction synovial cyst associated with atlanto-axial dislocation—case report. *Neurol Med Chir (Tokyo)* 1999;39:539–43.
7. Chang H, Park JB, Kim KW. Synovial cyst of the transverse ligament of the atlas in a patient with os odontoideum and atlantoaxial instability. *Spine* 2000;25:741–4.
8. Isono M, Ishii K, Kamida T, et al. Retro-odontoid soft tissue mass associated with atlantoaxial subluxation in an elderly patient: a case report. *Surg Neurol* 2001;55:223–7.
9. Zorzon M, Skrap M, Diodato S, et al. Cysts of the atlantoaxial joint: excellent long-term outcome after posterolateral surgical decompression. Report of two cases. *J Neurosurg* 2001;95:111–4.
10. Morio Y, Yoshioka T, Nagashima H, et al. Intradiscal synovial cyst communicating with the C1-C2 facet joints and subarachnoid space associated with rheumatoid atlantoaxial instability. *Spine* 2003;28:E492–5.
11. Sagiuchi T, Shimizu S, Tanaka R, et al. 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. Cecchi PC, Peltz MT, Rizzo P, et al. Conservative treatment of an atlantoaxial degenerative articular cyst: case report. *Spine J* 2008;8:687–90.
13. Marbacher S, Lukes A, Vajtai I, et al. Surgical approach for synovial cyst of the atlantoaxial joint: a case report and review of the literature. *Spine* 2009;34:E528–33.
14. Aksoy FG, Gomori JM. Symptomatic cervical synovial cyst associated with an os odontoideum diagnosed by magnetic resonance imaging: case report and review of the literature. *Spine* 2000;25:1300–2.
15. Aizawa T, Ozawa H, Kusakabe T, et al. C1/2 facet cyst revealed by facet joint arthrography. *J Orthop Sci* 2010;15:603–7.

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