Case report

Retro-odontoid pseudotumor concomitant with proximal adjacent cervical spondylotic myelopathy after multilevel anterior cervical discectomy and fusion

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ABSTRACT

Introduction and importance: Adjacent cervical spondylotic myelopathy (CSM) following anterior cervical discectomy and fusion (ACDF) presenting as a retro-odontoid pseudotumor (ROP) is uncommon. This consequence adversely affects hand function, causes gait imbalance and results in other disabilities for the patient. This report describes the successful surgical treatment of a patient with ROP associated with adjacent CSM following multilevel ACDF of the subaxial cervical vertebrae by performing posterior cervical decompression and fusion. Case presentation: A 60-year-old male presented with progressive, disabling cervical myelopathy. He had undergone ACDF C3-C7 for treatment of CSM 16 years ago and his symptoms had fully resolved. Magnetic resonance imaging (MRI) revealed severe cervical spinal cord compression caused by a retro-odontoid mass at the C1-C2 level with upper adjacent segment disease (ASD) of C1-C3. The patient received C1-C3 posterior cervical spinal fusion by C1 lateral mass C2 and C3 pedicle screw fixation and C1-C3 laminectomy. After the surgery, he was able to ambulate independently and the myelopathic symptoms were significantly improved at the 6 months follow-up.

Clinical discussion: Retro-odontoid pseudotumor concomitant with proximal ASD following ACDF is a rare occurrence. Both diagnosis and surgical management are challenging.

Conclusions: Posterior cervical decompression and fusion of C1-C3 is an effective option for treatment of severe cervical spinal cord compression by a retro-odontoid mass at the C1-C2 level combined with ASD after ACDF.

1. Introduction and importance

Anterior cervical discectomy and fusion (ACDF) is the standard treatment for cervical spondylotic myelopathy (CSM). ACDF for multilevel CSM is associated with a high incidence of adjacent segment disease (ASD). The revision surgery rate of ASD ranges from 2.1%–9.13% for single level ACDF, and 4.4%–10.7% for multilevel ACDF [1]. The upper cervical spine (C1-C2) has the lowest risk for ASD and appears in fewer reports [2]. Retro-odontoid pseudotumor (ROP) is a non-neoplastic lesion commonly associated with inflammatory conditions of unknown etiology which can mimic a malignant tumor. It is a chronic process seen in both inflammatory and non-inflammatory conditions. ROP associated with non-inflammatory (non-rheumatoid arthritis) disease is clinically rare [3,4]. In particular, non-inflammatory ROP-associated adjacent CSM after ACDF is seldom observed. This report presents a case of ROP-associated adjacent CSM after ACDF treated by posterior cervical decompression and fusion. This work has been reported in accordance with SCARE criteria [5].

2. Case presentation

A 60-year-old male presented with complaints of progressive upper extremity weakness and gait dysfunction for 3 months. He had undergone ACDF C3-C7 for treatment of CSM 16 years ago and neuromotor functions had fully recovered. He had no history of cervical spine trauma or rheumatoid arthritis. The patient did not smoke, drink alcohol, or use recreational drugs. Physical examination showed initial vital signs and mental status were normal. He had bilateral upper extremity weakness (motor power gr. III) and lower extremity weakness (gr. IV). Gait and balance were spastic and he was unable to ambulate without assistance.
Upper and lower extremity deep tendon reflexes were found hyperreflexic. Rectal examination found normal sphincter tone.

Radiographic imaging showed fused multilevel ACDF with plating C3-C7 and arthritic changes in C1-C3. (Fig. 1A). Magnetic resonance imaging (MRI) revealed severe degenerative joint disease (DJD) at C2-C3 with a retro-odontoid mass (Fig. 1B-C) at the C1-C2 level causing severe spinal cord compression. The retro-odontoid mass was a slightly T1-hypointense with a T2-hypointense soft tissue lesion. MRI results suggested a retro-odontoid pseudotumor as well as less likely diagnosis of gouty tophus, vertebral tumor or soft tissue tumor. Based on that, C1-C3 posterior cervical spinal fusion by performing C1 lateral mass-C2&C3 pedicle screws and C1-C3 laminectomy under general anesthesia was

Fig. 1. Pre-operative radiographic x-ray (A) Retro-odontoid mass (red asterisk) in MRI sagittal view (B) and axial view (C). Patient positioning with Mayfield cervical traction (D). Intraoperative O-arm-navigated C1-C3 screw insertion (E, F). Final constructs (G). Post-operative radiographic x-ray (H). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

Fig. 2. Gait and balance improved after surgery.
Table 1. Published case reports of adjacent segment disease (ASD) with retro-odontoid pseudotumor following cervical spondylosis surgery in the English-language literature.

| Author & year | Sex | Age | Primary disease | Primary surgery | Duration of ASD | Symptoms | Course of ASD | Final surgical treatment |
|---------------|-----|-----|----------------|----------------|----------------|----------|---------------|--------------------------|
| Matsumoto et al. [6], 2006 | Male | 76 | Cervical spondylosis | C3-C7 laminoplasty | 4 yrs. | Progressives quadriaparesis | Retro-odontoid pseudotumor | C1 laminectomy and posterior fusion from the occiput to C6 |
| Alomari et al. [7], 2019 | Male | 81 | Cervical spondylosis | C2 dome and C3-C7 laminoplasty | 10 yrs. | Gait and balance disturbance | Retro-odontoid pseudotumor | C1 laminectomy without instrument fixation |
| Miyata et al. [2], 2021 | Male | 77 | Cervical spondylosis | C3-C6 laminoplasty with C2/C7 partial laminectomies | 12 yrs. | Gait and balance disturbance | Retro-odontoid pseudotumor | Posterior fusion from the occiput to C2 without laminectomy |
| This case | Male | 60 | Cervical spondylosis | C3-C7 anterior cervical disectomy and fusion | 16 yrs. | Gait and balance disturbance | Retro-odontoid pseudotumor | C1-C3 laminectomy with posterior fusion from C1 to C3 |

selected. Cervical spine alignment was maintained with Mayfield cervical traction and the patient was placed in the prone position (Fig. 1D) for a posterior surgical approach. Intra-operative findings included severe osteoarthritis of C1-C3, severe spinal cord compression and solid bony fusion of C3-C7 vertebrae. C1 lateral mass-C2&C3 pedicle screws were inserted under O-arm navigation (Fig. 1E-F) with C1-C3 laminectomy by an experienced spine surgeon (TB) (Fig. 1G). Postoperative radiographic films showed an acceptable alignment and that rods and screws were in good position (Fig. 1H). The patient did not have postoperative complications and was discharged with gait and balance training rehabilitation program. At the follow-up visit 2 weeks after the operation, he was able to ambulate independently without using gait aids (Fig. 2). The patient cooperated with the rehabilitation program and came to all scheduled follow-up appointments. At the 6-month follow-up, his condition had significantly improved. The patient was highly satisfied with the outcome of the treatment and was able to return to work normally at his former occupation.

3. Clinical discussion

The study subject is the ROP concomitant with proximal adjacent CSM after multilevel ACDF, which is a very rare condition. A review of the literature and case reports showed very few cases of ROP following cervical spondylosis surgery (Table 1). Three studies [2,6,7] each reported on a single case of proximal ASD with retro-odontoid pseudotumor following cervical spondylosis surgery. All cases received surgical treatment, but using different techniques. Only one, Alomari et al. [7], reported performing a laminectomy without instrumentiation. However, we suggest using instrumentation to reduce the risk of iatrogenic instability following a laminectomy.

ROP has been categorized by Tanaka et al. [8] and classified into 3 types according to the etiology and MRI findings. Type 1 has the highest incidence, with MRI identifying hypo- or isointense on T1-weighted lesions and heterogeneous (hyper- and hypointense) in T2-weighted lesions caused by atlantoaxial subluxation associated with rheumatoid arthritis (RA). Type 2 has been reported to be rare, with MRI findings similar to type 1, but resulting from ankylosis, ossification of the anterior longitudinal ligament (OALL) or spondylosis of the cervical spine [8]. Type 3 is also rare, with an incidence identical to type 2. ROP has found hypo- or isointense in T1-weighted lesions and hypointense C2-C3 disc herniation, contiguous with retro-odontoid space in T2-weighted images.

In the present case, we report on a ROP in a non-rheumatoid arthritis patient with cervical myelopathy. The MRI finding was classified as type 2 of Tanaka’s classification. Tanaka et al. [8] recommended posterior C1-C2 fixation and laminectomy for surgical management in ROP types 1 and 2 [8]. However, this case was a type 2 ROP concomitant with ASD after multilevel ACDF, making it extremely rare and challenging for preoperative surgical treatment. The author chose C1-C3 posterior fixation and spinal cord decompression by performing cervical laminectomy because the upper adjacent segment along C1-C3 and the rest of cervical spine was rigid due to previous anterior fusion of C3-C7. We proposed a fixation technique using C1 lateral mass-C2&C3 pedicle screws and C1-C3 laminectomy.

4. Conclusions

In severe cervical spinal cord compression by the retro-odontoid mass at the C1–C2 level combined with C2-C3 ASD after ACDF, posterior cervical decompression and fusion C1-C3 is an effective treatment and might be an option in this condition.

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Ethical approval

This case report was approved by the Institutional Review Board, Faculty of Medicine, Chiang Mai University.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in Chief of this journal upon request.

Registration of research studies

None.

Guarantor

Torphong Bunmaprasert, MD, Associate Professor.

CRediT authorship contribution statement

Wongthawat Liawrungrueang: Conceptualization, Methodology, Visualization, Writing -original draft.
Waroon Tantivorawit: Resources, Data curation.
Nantawit Sugandhavesa: Resources, Data curation.
Torphong Bunmaprasert: Data curation, Writing - review & editing, Supervision.

Declaration of competing interest

None.
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