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Surgical Strategy for a Retro-Odontoid Pseudotumor

Katsunori Fukutake, Hiroshi Takahshi*, Yuichirou Yokoyama, Keiji Hasegawa, ShintaroTsuge, Kazumasa Nakamura, RyoTakamatsu, Akihito Wada and Yasuaki Iida

Abstract

Retro-odontoid pseudotumors that are not associated with rheumatoid arthritis or hemodialysis are clinically rare. A pseudotumor is a non-neoplastic mass in the retro-odontoid region that can occur in elderly people. Retro-odontoid pseudotumors are regarded as non-neoplastic masses that arise as a result of instability of the atlantoaxial joint. However, Retro-odontoid pseudotumor has occasionally been reported without instability on dynamic radiography. In general, atlantoaxial or occipito-cervical fusion is selected for patients with instability. For patients without instability, favorable outcomes have been reported following laminectomy of the atlas. We encountered 3 patients with Retro-odontoid pseudotumor and treated them surgically. In this study, we investigated the validity of the previously discussed surgical therapies, based on our patients. Laminectomy of the atlas did not reduce the pseudotumor size, and improvement of clinical symptoms was poor. In contrast, in the patient treated with occipito-cervical fusion, the pseudotumor disappeared rapidly, and the clinical symptoms improved. If the cause of this disease is instability or overload of the atlantoaxial joint, it is desirable to concomitantly apply fusion even though instability has not been observed on preoperative dynamic radiography, and, anatomically, occipito-cervical fusion should be performed.

Keywords

Retro-odontoid pseudotumor; Surgery; Occipito-cervical fusion

Introduction

Retro-odontoid pseudotumors that are not associated with rheumatoid arthritis or hemodialysis are clinically rare. Recently, mass lesions around the axis that are unassociated with underlying disease have also been reported. Retro-odontoid pseudotumor is regarded as a non-neoplastic mass that arises as a result of instability of the atlantoaxial joint. However, cases of Retro-odontoid pseudotumors without instability on dynamic radiography have been occasionally reported.

While atlantoaxial fusion or occipitocervical fusion are generally selected for cases with instability, favorable outcomes of laminectomy of the atlas have been reported in cases without instability. We encountered 3 patients with surgically treated retro-odontoid pseudotumors. In this study, we investigated the validity of the previously reported surgical treatments that have been discussed, based on the outcomes of surgical procedures in 48 cases that have been reported in Japan, including our 3 patients.

Subjects and Methods

The subjects were 3 patients who underwent surgery at our hospital and 45 patients that were previously reported in Japan, for a total of 48 cases. There were 38 male and 10 female patients, and the mean age at the time of surgery was 75.6 years. Instability of the atlantoaxial joint was evaluated using dynamic plain X-ray imaging. Instability was noted in 26 patients before surgery. Regarding the diagnostic criteria for instability, the atlanto-dental interval (ADI) was measured in the flexion, neutral, and extension positions, and an ADI ≥ 3 mm in anteflexion was diagnosed as instability.

Case Reports

Case 1: An 81-year-old man

Chief complaints: Neck pain and disturbance of fine motion of fingers. Neck pain had developed 3 years earlier, disturbance of fine motion of fingers appeared one year earlier, and the patient had undergone MRI at a clinic.

A retroodontoid tumorous lesion was noted, and the course was followed, but gait disorder rapidly became aggravated, and the patient was referred to our hospital for surgery. Upon admission, no muscle weakness was noted in the four limbs, but bilateral disturbance of fine motion of fingers was noted in the fingers. Deep tendon reflexes were enhanced in the four limbs, and spastic gait was observed. Bulbar paralysis and bladder and rectal disturbances were absent.

On plain X-ray imaging, spondylosis was observed, and ossification of the anterior longitudinal ligament (OALL) was also noted in the second cervical vertebra and lower vertebrae. Arthropathic changes in the atlantoaxial joint were mild, and the ADI on dynamic imaging was 3.8 mm in flexion, 4.0 mm in the neutral position, and 4.2 mm in extension, showing instability. MRI demonstrated a tumorous lesion showing low intensity on T1-weighted imaging and mixed low and high intensities on T2-weighted imaging, and the spinal cord was severely excluded toward the left side. The dural canal was excluded at the levels of C4/5 and C5/6 (Figure 1a).

The diagnosis was retro-odontoid pseudotumor with instability, but fusion was not applied in consideration of the age and general condition, and laminoplasty of C4 and C5 was performed in addition to laminectomy of the atlas. The symptoms remitted immediately after surgery but gradually recurred. Four years after surgery, symptoms have recurred, similar to those before surgery. The pseudotumor size has not changed on MRI (Figure 1b).

Case 2: An 80-year-old man

Chief complaints: Bilateral upper limb numbness, disturbance offine motion of fingers, and gait disorder. The chief complaints developed one year earlier, and the patient visited a physician. A tumorous lesion posterior to the axis was found, and the course was followed. Gait disorder gradually became aggravated over the course of one month, and the patient was referred to our hospital for surgery. Upon admission, no muscle weakness was noted in the four

***Corresponding author:** Hiroshi Takahshi, Toho University School of Medicine, Department of Orthopaedic Surgery, Japan, Tel: 81-3-3762-4151; Fax: 81-3-3763-7539; E-mail:drkan@med.toho-u.ac.jp

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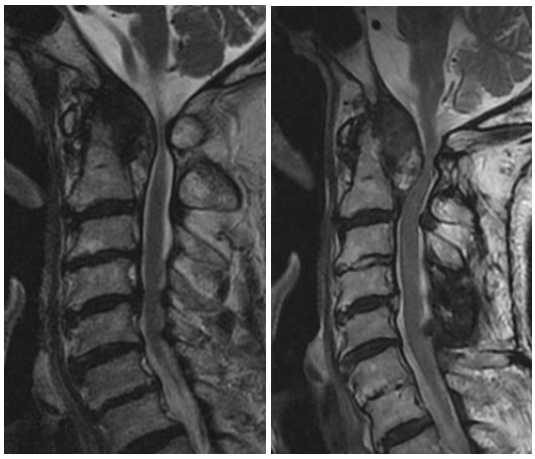

Figure 1: a: Preoperative T2 W.I., b: T2 W.I. 4 years after surgery

A tumorous lesion showing low intensity on T1-weighed imaging and mixed low and high intensities on T2-weighed imaging was present posterior to the odontoid. MRI 4 years after the surgery showed no change in the pseudotumor size.

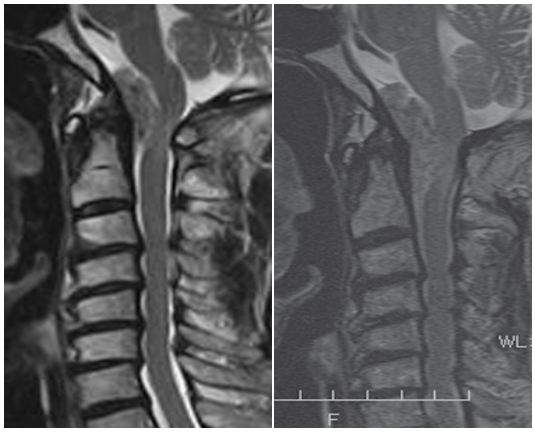

Figure 2: a: Preoperative T2 W.I., b: T2 W.I. 3 years after surgery

A tumorous lesion showing low intensity on T1-weighed imaging and mixed low and high intensities on T2-weighed imaging was present posterior to the odontoid, and the spinal cord was severely excluded leftward. MRI 3 years after the surgery showed that the pseudotumor was enlargened.

limbs, but bilateral peripheral numbness was observed in the four limbs, the deep tendon reflex was enhanced in the four limbs, and the Hoffman reflex was positive. Bulbar paralysis and bladder and rectal disturbances were absent.

On plain X-ray imaging, no arthropathic changes were observed in the atlantoaxial joint. On dynamic imaging, the ADI was 2.8 mm in flexion, 2.6 mm in the neutral position, and 2.6 mm in extension, showing no instability. MRI demonstrated a tumorous lesion showing low intensity on T1-weighted imaging and mixed low and high intensities on T2-weighted imaging, and the spinal cord was severely excluded toward the left side (Figure 2a).

The patient was diagnosed with retro-odontoid pseudotumor without instability, and laminectomy of the atlas was performed. After surgery, the neurological symptoms improved temporarily, but gradually recurred within one year. Three years after the surgery,

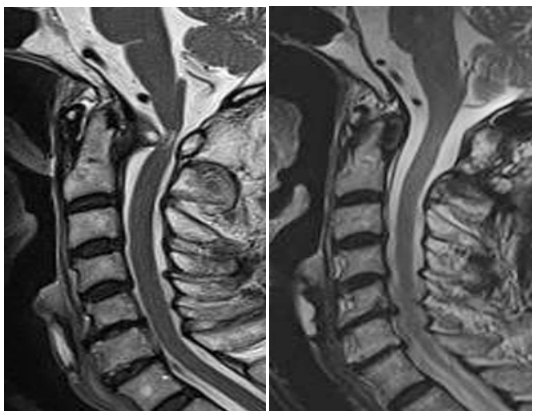

Figure 3: a: Preoperative T2 W.I., b: T2 W.I. 5 months after surgery

A tumorous lesion showing low intensity on T1-weighed imaging and mixed low- and high-intensities on T2-weighed imaging was evident posterior to the odontoid, and the spinal cord was severely excluded. MRI at 5 months after the surgery showed that the pseudotumor had mostly disappeared.

symptoms are similar to those before surgery, and MRI shows enlargement of the pseudotumor (Figure 2b).

Case 3: A 75-year-old man

Chief complaints: Four limb numbness. Left side-dominant four limb numbness appeared 3 months earlier, and the patient underwent MRI at a clinic. A tumorous lesion posterior to the axis was found, and the patient was referred to our hospital for close examination and treatment.

Upon admission, disturbance of fine motion of fingers of the fingers and muscle weakness of the left toes were noted. The deep tendon reflex was enhanced in the four limbs, but bulbar paralysis and bladder and rectal disturbances were absent.

On plain X-ray imaging, no arthropathic changes were noted in the atlantoaxial joint. The ADI on dynamic imaging was 6.1 mm in flexion, 3.0 mm in the neutral position, and 2.2 mm in extension, showing instability. On MRI, a tumorous lesion was detected showing a low intensity on T1-weighted imaging and mixed low and high intensities on T2-weighted imaging, and the spinal cord was severely excluded (Figures 3a and 3b). The patient was diagnosed with retro-odontoid pseudotumor with instability, and occipito-cervical fusion was applied in addition to laminectomy of the atlas. The symptoms improved rapidly after surgery. One year after surgery, only mild numbness remains in the fingers (Figure 4). On MRI, the pseudotumor shrunk after 3 months and mostly disappeared within 5 months (Figure 3b).

Discussion

A pseudotumor is a non-neoplastic mass in the retro-odontoid region that occurs in elderly people. Regarding the pathology of pseudotumors, Sze et al. [1] investigated 3 patients. They reported that stimulation by long-term persistence of atlantoaxial subluxation was involved in the mass formation, and that the true condition was the accumulation of fibrous granulation tissue on histopathological examination. Crockard et al. [2] reported 5 patients who developed retro-odontoid pseudotumor, despite the absence of instability of the atlantoaxial joint, and in the absence of inflammatory cells in the tissue, and showed that the tumor was comprised of an amorphous,

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Figure 4: Lateral plain X-ray image one year after surgery Occipitocervical fusion was applied. Bone union was favorable. The patient had no complaint of limitation of the range of motion or neck pain.

acellular material containing fibrous tissue and cartilage, assumed to have resulted from repeated micro rupture and repair of the transverse ligament of the atlas. Yoshida et al. [3] reported 5 patients with retro-odontoid pseudotumor accompanying atlantoaxial joint instability of unknown cause, in which the histopathological finding was degenerated fibrous tissue, partially accompanied by fibrin deposition, and many chondrocyte-like cells, but in the absence of inflammatory cells, which was diagnosed as fibrous cartilage tissue. One patient was autopsied, and the mass was found to be continuous to the retro-odontoid cruciate ligament component.

Chikuda et al. [4] investigated 10 patients with retro-odontoid pseudotumor. Setting the criterion of atlantoaxial joint instability at an atlantodental interval (ADI)>4 mm, instability was present in only 2 of the 10 patients; thus, instability was absent in 8 patients. They reported that atlantoaxial joint instability was not always present in pseudotumor formation, and that mobility of the cervical vertebrae below the axis was reduced due to the progression of OALL in these patients. This condition produced excess load on the atlantoaxial joint, thus contributing to the pseudotumor formation.

Regarding the treatment of pseudotumor, total excision of the mass was previously considered ideal because the pathology was clear, and total excision has been performed via the transoral approach, as reported by Crockard et al. [2]. However, 2 of their 5 patients died of pneumonia after surgery. Considering the surgical stress, the duration of bed rest after surgery, and increased atlantoaxial joint instability after surgery, this treatment may not be safe.

Yukiyama et al. [5] reported the outcomes of laminectomy of the atlas in 2 elderly patients older than 80 years old, one with instability and the other without instability. The mass size was not changed after surgery in either patient, but the spinal cord symptoms were improved, and the surgery was minimally-invasive and effective. Sze et al. [1] also reported 3 patients treated with laminectomy of the atlas.

It has recently been reported that the mass shrunk or disappeared over time on MRI after application of atlantoaxial [6,7] or occipitocervical fusion [4,8], without direct excision of the mass. Thus, Fukutake et al. [9] reported the surgical outcomes of 48 patients with retro-odontoid pseudotumor at the 48th Meeting of the Japan Medical Society of Spinal Cord Lesion. The surgical procedure was laminectomy of the atlas in 19 patients, atlantoaxial fusion in 4, and occipitocervical fusion in 25. Clinical symptoms were improved in 42 of the 48 patients, but not in 6 patients. Based upon the surgical procedure, improvement was obtained in 14 of the 19 patients treated with laminectomy of the atlas. Of the 4 patients treated with atlantoaxial fusion, symptoms were improved in 3, whereas clinical symptoms were improved in all of the 25 patients treated with occipitocervical fusion. Occipitocervical fusion was additionally applied to 2 of the 5 patients in whom laminectomy of the atlas did not improve symptoms, and led to improvement of the symptoms. Regarding the size of the retro-odontoid pseudotumor, it was reduced in 2 of the 19 patients treated with laminectomy of the atlas, 3 of the 4 patients treated with atlantoaxial fusion. Of the 25 patients treated with occipitocervical fusion, the tumor size decreased in 14 and the tumor disappeared in 9. These findings demonstrated that fusion, particularly occipitocervical fusion, is far superior to laminectomy of the atlas in improvement of the clinical symptoms and reduction in the size or resolution of the pseudotumor.

Of our 3 patients, 2 were treated with laminectomy of the atlas alone and one was treated with occipitocervical fusion + laminectomy of the atlas. In the 2 patients treated with laminectomy of the atlas alone, symptoms were improved temporarily after surgery but gradually aggravated over time. In contrast, in the patient in whom fusion was also applied, the mass disappeared with time on MRI, and favorable improvement of the symptoms was achieved.

Evaluation of the surgical outcomes in elderly patients is difficult because age-related phenomena must also be considered. As observed in our patients, improvement was noted immediately after laminectomy of the atlas, but the outcome became aggravated after 3-4 years.

Anatomically, the ligament posterior to the odontoid is termed the cruciate ligament, and it is comprised of the transverse ligament of the atlas, which is the transverse portion, and the longitudinal region extending from the foramen magnum to the second cervical vertebra body. Considering the cause of the mass, occipitocervical fusion is reasonable.

Based on the above findings, in the treatment method of this disease, laminectomy of the atlas may contribute to temporary improvement of the symptoms, but these symptoms may become aggravated over long-term observation. When the addition of fusion is planned, occipitocervical fusion should be selected, rather than atlantoaxial fusion. However, this disease frequently develops in the elderly and sufficient explanation of the disease condition, pathology, and postoperative treatment and patient consent are necessary.

Conclusion

We encountered 3 patients with retro-odontoid pseudotumors. Laminectomy of the atlas did not reduce the pseudotumor size, and improvement of clinical symptoms was poor. Occipitocervical fusion rapidly resolved the pseudotumor and improvement of clinical symptoms was favorable. When the cause of this disease is atlantoaxial joint instability or overload, it is desirable to concomitantly apply fusion, even when instability is not observed on preoperative dynamic imaging, and anatomically, occipitocervical fusion should be performed.

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Author Affiliation

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Toho University School of Medicine, Department of Orthopaedic Surgery, Japan