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Case Report

Salvage Surgery for Symptomatic Recurrence of Retro-Odontoid Pseudotumor after a C1 Laminectomy

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We provide the first report of successful salvage surgery for a post-C1 laminectomy symptomatic recurrence of a retro-odontoid pseudotumor (ROP) that caused myelopathy. The 72-year-old Japanese woman presented with an ROP causing symptomatic cervical myelopathy. With ultrasonography support, we performed the enucleation of the ROP via a transdural approach and fusion surgery for the recurrence of the mass. At the final observation 2-year post-surgery, MRI demonstrated the mass's regression and spinal cord decompression, and the patient's symptoms had improved. Our strategy is an effective option for a symptomatic recurrence of ROP.

Key words: recurrent retro-odontoid pseudotumor, salvage surgery, transdural resection, C1 laminectomy, ultrasonography

A retro-odontoid pseudotumor (ROP) at the craniocervical junction is a rare condition in patients without inflammatory diseases such as rheumatoid arthritis and those undergoing long-term hemodialysis [1,2]. There are multiple surgical options for this pathology, including transoral resection [3] or posterior fusion [4-11], C1 laminectomy [12-14], far lateral resection [15], and transdural resection [16-19]. Posterior fusion is a reasonable surgical strategy for an ROP with atlantoaxial subluxation [4-11]. Decompression surgery as a C1 laminectomy is also selected in cases without marked atlantoaxial instability or high-risk general conditions; this method reportedly facilitates relatively good clinical outcomes [12-14]. However, decompression surgery has a limited effect on the regression of a pseudotumor, and it occasionally causes recurrence [13,20,21], and cases with a symptomatic recurrence of ROP after a C1 laminectomy have been reported [21]. To the best of our knowledge, surgical strategies for the symptomatic recurrence of an ROP have not been extensively explored in the literature. We provide a first report of a salvage surgery for the symptomatic recurrence of an ROP causing myelopathy in a patient without atlantoaxial instability but exhibiting progressive neurological deterioration after a C1 laminectomy.

Case Presentation

The patient was a 72-year-old Japanese woman who had developed numbness in her upper and lower extremities 6 years ago. At that time, she was diagnosed with myelopathy due to an ROP and underwent a C1 laminectomy as a decompressive surgery at another hospital (Fig. 1). Although her complaint had resolved completely 2 years after that surgery (Fig. 2), numbness reappeared in the upper and lower extremities. She had visited our institute 3 years ago because myelopathic

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symptoms (gait disturbance and fine movement disturbance in the left hand) were exacerbated. Her past medical history was not significant.

The neurological examination on her first visit to our institute revealed abnormal reflexes, including the pathological plantar reflex. A manual muscle test revealed no weakness, but numbness and sensory loss were detected in the upper and lower extremities. The patient complained of clumsiness in the hands and gait disturbance; no bladder dysfunction was present. Subacute progressive deterioration of neurologic symptoms was observed. The Japanese Orthopedic Association (JOA) score for cervical myelopathy was 10 out of a maximum of 17 points.

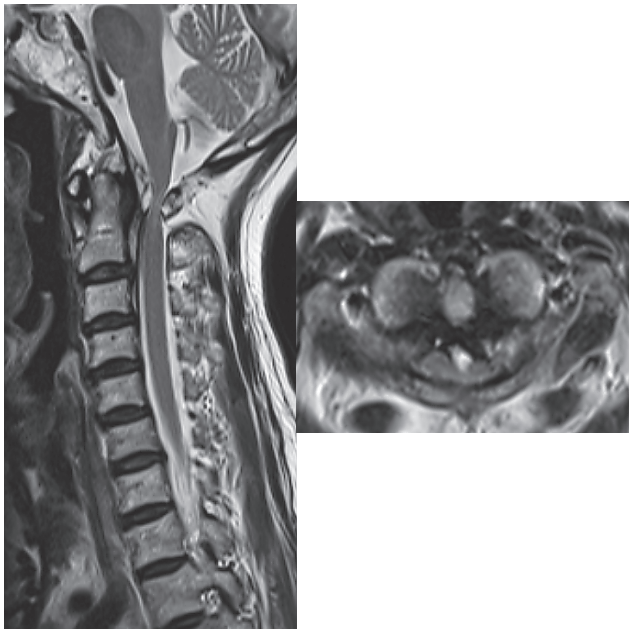

Fig. 1 Sagittal (*left*) and axial (*right*) views of T2-weighted MRI before the patient's primary surgery revealing a retro-odontoid mass with a cyst compressing the spinal cord.

Fig. 2 Sagittal (*left*) and axial (*right*) views of T2-weighted MRI at 1 year postoperatively showing decompression of the spinal cord.

Plain radiography showed mild osteoarthritis at the C1/2 joint with sclerotic changes in the odontoid process and mild degenerative spondylolisthesis in multiple cervical segments. Dynamic radiography exhibited no instability at the C1/2 level (Fig. 3). The atlas-dens interval was 2 mm on cervical flexion. Magnetic resonance imaging (MRI) demonstrated a retro-odontoid mass accompanied by a multilocular cyst; these were severely compressing the patient's spinal cord (Fig. 4A, B). A high-intensity area within the spinal cord was observed on the T2-weighted image. Plain computed tomography (CT) showed the postoperative C1 laminectomy (Fig. 4B).

With the use of the posterior approach and under multimodal intraoperative spinal cord monitoring, dura exposure was achieved by conducting a C2 French-door laminoplasty. We first tried to detach the lateral side of the dura from surrounding tissue but could not, as the anterolateral aspect of the dura and surrounding soft tissue were firmly adhered. We thus chose a posterolateral transdural approach to the mass of ROP (Fig. 5A). After the dura was opened, the spinal cord was gently retracted with the denticulate ligament. After the positional relationship between the mass and spinal cord was confirmed with an ultrasonographic device (LOGIC e Premium®, GE Healthcare Japan, Tokyo) (Fig. 5B), a straight and longitudinal approx. 1-cm incision was made in the anterior part of the dura.

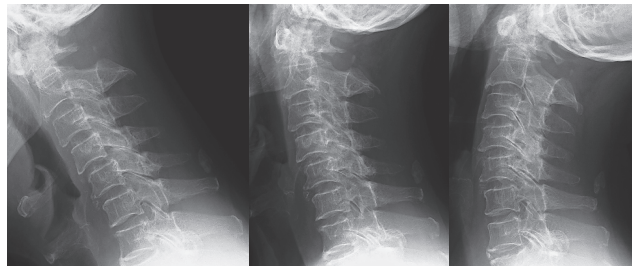

Fig. 3 Dynamic radiography showing no atlantoaxial subluxation.

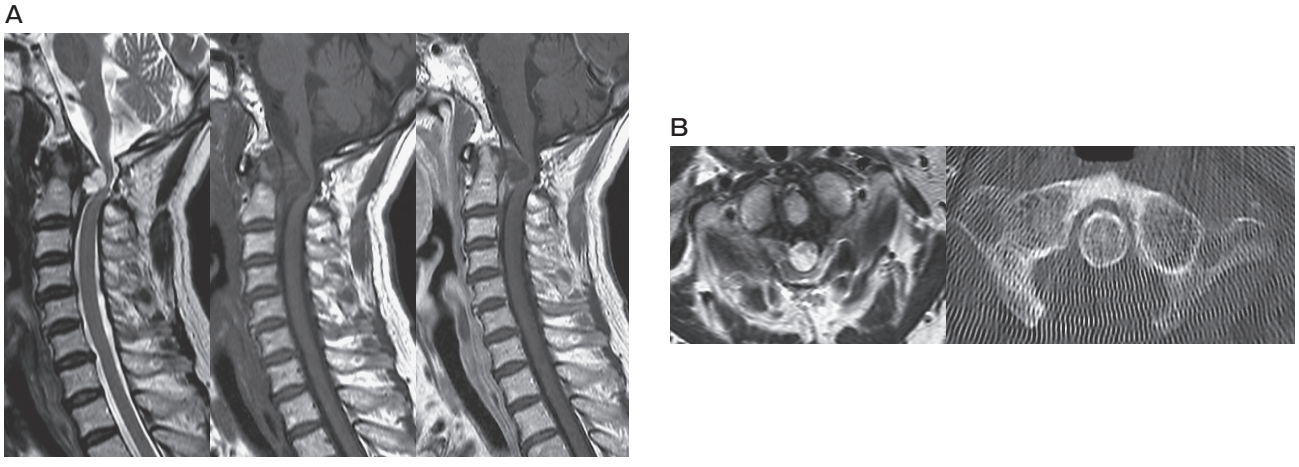

Fig. 4 **A**, Sagittal view of T2- (*left*) and T1-weighted MRI (*middle*) before the revision surgery showing the pseudotumor with a multilocular cyst compressing the spinal cord and a high-intensity area within the spinal cord. T1-weighted MRI with gadolinium (*right*) shows peripheral enhancement of the mass; **B**, Axial view of T2-weighted MRI before the revision surgery (*left*) showing the pseudotumor with cyst severely compressing the spinal cord. Axial view of the plain CT (*right*) showing a defect of the arch of the atlas after the C1 laminectomy.

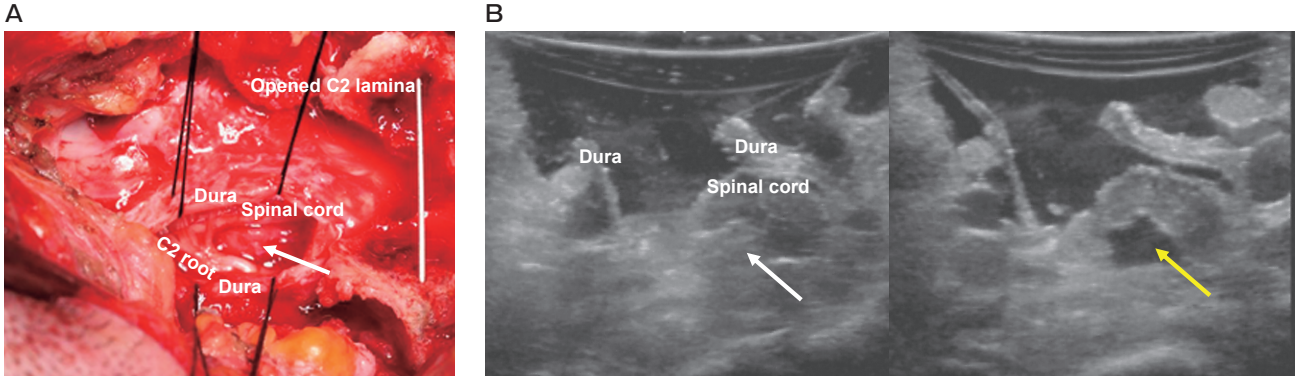

Fig. 5 **A**, Intraoperative photograph obtained after the dorsal dura was incised, showing a torus of the retro-odontoid mass (*white arrow*); **B**, Intraoperative ultrasonographic images obtained before the resection of the retro-odontoid lesion (*left*) showing the compressed spinal cord due to the retro-odontoid mass (*white arrow*) and a large space on the ventral side of the spinal cord after the mass was resected (*yellow arrow*) and the still decompressed spinal cord (*right*).

The vascular development was poor, and no granular tissue was observed. After the whitish erupted pseudotumor was removed along with the cyst, the mass located in front of the spinal cord was dissected under ultrasonography (Fig. 5B). Spinal cord decompression was sufficiently achieved intraoperatively with ultrasonography support. After the dura was closed, C1/2 fusion surgery using C1 lateral mass screws and C2 pedicle screws was completed. Finally, decortication, resection of the cartilaginous surface with a curette, and bone grafting were conducted at the C1/2 facet joint.

A microscopic examination revealed that the resected material contained fibrocartilaginous tissue with degenerative necrosis; however, inflammatory cell infiltrates and signs of neoplasm were not observed. The histopathological findings confirmed the diagnosis of ROP.

Immediately after the surgery, the patient recovered from the fine-movement disorders in the upper extremities. Gradual improvement in her gait disturbance and numbness in the upper and lower extremities was observed, and the patient was thus ambulant without a cane at discharge. At the final follow-up 2 years after the surgery, CT showed bone fusion at the C1/2 facet, and MRI revealed decompression of the spinal cord;

the absence of recurrence of the ROP mass was confirmed (Fig. 6). There has been no evidence of postoperative adhesive arachnoiditis. The patient's JOA score had improved from 10 to 16, with an 86% recovery rate.

This study was approved by our institutional review board before its initiation (No. 3266) and was conducted in accordance with the Declaration of Helsinki for medical research involving human subjects. The patient provided written informed consent for the publication of her case and associated data.

Ethical approval. This study was performed in accord with the principles of the Declaration of Helsinki and was conducted with the approval of the Research Ethics Committee of Wakayama Medical University (No. 3266).

Discussion

We have described a successful salvage surgery for myelopathy due to the recurrence of an ROP; the surgery involved a transdural resection with ultrasonography support. The basic strategy for the surgical treatment of a symptomatic ROP has been described, and given that the pathomechanism of ROP formation includes chronic mechanical stress on the atlantoaxial joint [1, 2], posterior fusion is theoretically reasonable. Many authors have stated that posterior C1-C2 fusion regresses the retro-odontoid soft tissue mass associated with atlantoaxial subluxation and improves myelopathy symptoms [4-11]. Chikuda *et al.* demonstrated that an ROP mass is caused by chronic mechanical stress as an adjacent segmental disorder associated with the fused spine, such as diffuse idiopathic skeletal hyperostosis and ossification of the posterior longitudinal ligament [10]. A longer cranial-cervical fusion may thus be selected, even in the absence of obvious instability at the atlantoaxial joint [10, 11].

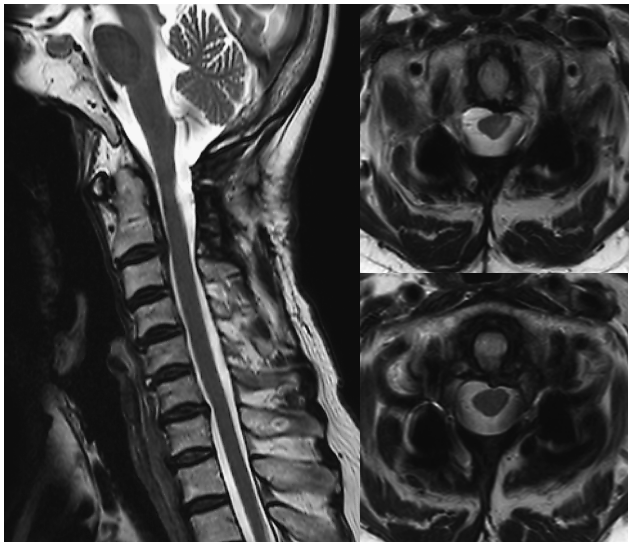

Fig. 6 Postoperative sagittal and axial T2-weighted MRI obtained 2 years after the revision surgery (posterior resection and fusion) showing the decompressed spinal cord and no recurrence of the retro-odontoid pseudotumor lesion.

However, decompressive surgery alone is also often selected in patients who are elderly, at high risk for surgery, and/or cannot tolerate a postoperative reduced range of motion of the cervical spine. A C1 laminectomy alone has led to the improvement of myelopathy symptoms and the regression of the retro-odontoid soft tissue mass, which indicates some effectiveness of posterior decompression surgery [12-14]. Conversely, some reports have described limitations of decompression surgery for a symptomatic ROP, even in cases without atlantoaxial instability. Kobayashi *et al.* observed significantly less regression of the pseudotumor mass after decompression surgery compared to after fusion surgery [20]. Cases of a symptomatic recurrence of an ROP after a C1 laminectomy have also been reported [21].

The optimal surgical strategy for myelopathy symptoms due to a recurrent ROP has not been thoroughly discussed in the literature. For the present patient, we selected posterior fusion with a direct resection of the pseudotumor because the large ROP mass lesion with a cyst had caused a subacute progressive deterioration of the patient's neurological symptoms due to severe compression of the spinal cord. However, we chose the posterolateral transdural approach to the ROP mass because it was difficult to reach the ROP mass via an extradural approach due to the strong adhesion at the lateral side of the dura. Regarding the surgical indications for a transdural resection of ROP, Fujiwara *et al.* noted that an ROP can cause myelopathy and that such an ROP mass lesion is large enough to require direct resection [16]. We also performed a C1/2 fusion surgery for our patient with the use of C1 lateral mass screws and C2 pedicle screws, which have been shown to maintain reliable stability for atlantoaxial instability [22-24].

There are some recent reports describing a transdural resection of an ROP [16-19], but this method has not been adapted for a symptomatic recurrence of an ROP. We were concerned about the adhesions between

our patient's ventral dura and the pseudotumor mass, as Fujiwara *et al.* described [16]. Schomacher *et al.* concluded that a posterior transdural resection is contraindicated when preoperative CT indicates that the mass includes calcification, because it is challenging to perform detachment due to severe adhesion [19]. This has been the primary concern in transdural resections for symptomatic recurrences of ROPs.

To address this concern, our intraoperative use of ultrasonography allowed us to accurately assess the degree of adhesions. Another advantage of using ultrasonography in the surgical field is that its use clarifies the positional relationship between the spinal cord and ROP mass intraoperatively in real time. A third advantage is that the endpoint of decompression can be easily determined with ultrasonography. Thus, by using intraoperative ultrasound guidance in the present transdural resection, we were able to adequately and safely resect the ROP mass ventral to the patient's spinal cord while avoiding spinal cord injury. This method may be useful in the reoperation of patients suffering from a symptomatic recurrence of ROP. It is important to note that there are serious risks associated with the transdural resection approach; when the dura is opened, it is at risk of complicated adhesive arachnoiditis, which may cause spinal syrinx and myelopathy. We therefore conducted the present revision surgery very meticulously.

Conclusion

Salvage surgery for myelopathy due to the recurrence of an ROP after a Cl laminectomy was successfully performed via a transdural approach and with the support of ultrasonography. The intraoperative ultrasonography allowed an easy understanding of the position of the ROP and spinal cord, the endpoints of decompression, and the adhesions around the patient's spinal cord in real time. The surgical method described herein may thus be useful in the treatment of symptoms due to recurrent ROP.

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