Cervical Myelopathy with Retroodontoid Pseudotumor Caused by Atlantoaxial Rotatory Fixation and Senile Tremor

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Objective: A rare case of retroodontoid pseudotumor caused by the combination of atlantoaxial rotatory fixation and senile tremor is reported.
Method: A 63-year-old man was treated for cervical myelopathy with retroodontoid pseudotumor caused by atlantoaxial rotatory fixation and senile tremor. He had an 18-year history of torticollis and a 7-year history of tremor. Magnetic resonance imaging revealed a retroodontoid pseudotumor compressing the spinal cord. Computed tomography showed an atlantoaxial rotatory fixation and osteoarthritis of the lateral facets. Resection of the posterior arch of the atlas and a posterior occiput-to-axis arthrodesis were performed with an autologous bone graft. His neurological condition improved, and the retroodontoid pseudotumor disappeared at 3-year follow up.
Conclusions: Frequent rotation of the head by the senile tremor caused friction of the lateral atlantoaxial facet with the development of osteoarthritis. This long-term mechanical stress was thought to cause the soft tissue hypertrophy around the lateral facet. Although spontaneous resolution of the pseudotumor after fixation of the unstable segment has been reported as the treatment of choice, decompression with occipitoaxial fusion was selected in the current case because the patient's neurological deficit was severe and progressive because of a remarkable canal stenosis at level C1/2.

Key words: retroodontoid pseudotumor, atlantoaxial rotatory fixation, senile tremor, cervical myelopathy

INTRODUCTION

Retroodontoid pseudotumors are caused by inflammatory granulation or reactive soft tissue hypertrophy from chronic atlantoaxial subluxation of inflammatory or traumatic origin [1, 2]. Rheumatoid arthritis is the most frequent cause of retroodontoid pseudotumors [3, 4]. Mild tremor is a common accompaniment of normal aging. This tremor in older adults has been labeled "senile tremor" or "benign tremor" of the elderly [5, 6]. It is generally asymptomatic and does not require treatment. The disorder of the atlantoaxial joint, contraction of sternocleidomastoid muscle, etc. have been reported to be the cause of torticollis [7, 8]. A rare case of cervical myelopathy with retroodontoid pseudotumor is presented, which we suspected was caused by a combination of senile tremor and torticollis arising from atlantoaxial rotatory fixation.

CASE REPORT

A 63-year-old male with an 18-year history of torticollis began feeling numbness in his right upper extremity in September 2005. His numbness progressed, with the gradual onset of clumsiness. He was admitted to our institute in February 2006. His head was tilted to the left and his neck was rotated toward the right. He exhibited a rapid rhythmic movement of the anterior upper neck at rest. This tremor, which his family first noticed 7 years ago, abated whenever he was speaking or eating. Examination showed that he had marked spastic tetraparesis, with muscle power of at least Grade 4 in the right upper and lower extremities. Sensory disturbance in a distal (stocking-glove) distribution and bilateral clumsiness of the hands were observed. There was hyperreflexia throughout the upper and lower extremities. He did not show any evidences of rheumatoid arthritis that met the criteria of the American Rheumatism Association.

Plain radiograms showed the cervical spine tilted to the left and severe spondylosis on the left side (Fig. 1a). The posterior atlantodental interval was fixed at 2 mm without any dynamic increase. Computed tomography revealed atlantoaxial rotatory fixation, which we classified as Type 1, as per Fielding and Hawkins [7] (Fig. 1b, c). Magnetic resonance imaging (MRI) showed a large and smooth mass behind the odontoid process compressing the spinal cord. The density of the mass was intermediate on T1-weighted images and low on T2-weighted images. There was no apparent enhancement on gadolinium-enhanced images (Fig. 2).

A posterior surgical approach was used. The posterior arch of the atlas was resected, and a posterior occiput-to-axis arthrodesis was performed with autologous bone graft.

Within a week of the surgery, the patient showed remarkable improvement of his clumsiness and spasticity. Three months after surgery, he was able to resume his job as a barber. On MRI at 3 years follow up, the

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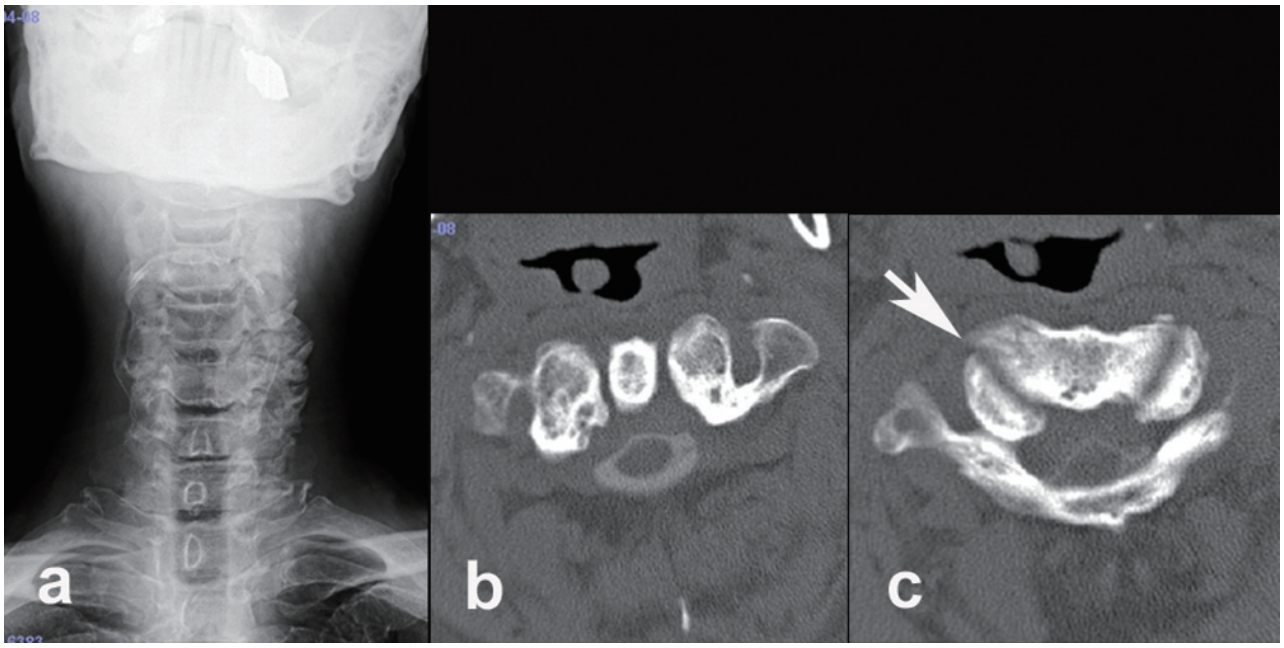

Fig. 1 a. Plain radiogram showing the torticollis and spondylosis on the left side. b, c. Preoperative computed tomography (b: at the level of middle of C1 lateral mass, c: at the level of base of odontoid process) showing atlantoaxial rotatory fixation and osteoarthritis of facet joint (Arrow).

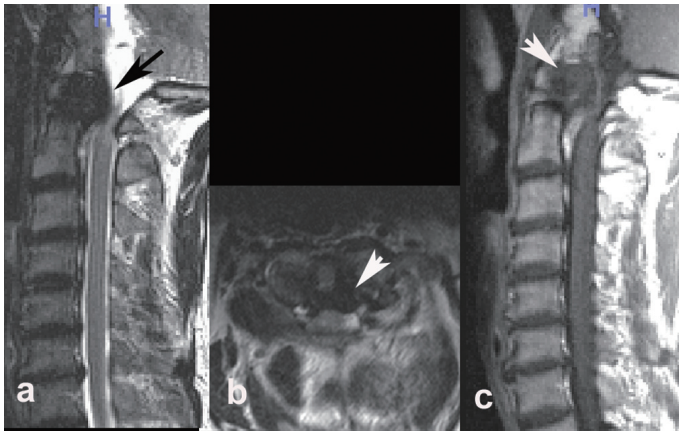

Fig. 2 Preoperative T2-weighted MRI (a: sagittal image, b: axial image) showing the retroodontoid pseudotumor compressing the spinal cord. c: Preoperative MRI (T1-weighted after administration of gadolinium) showing no enhancement.

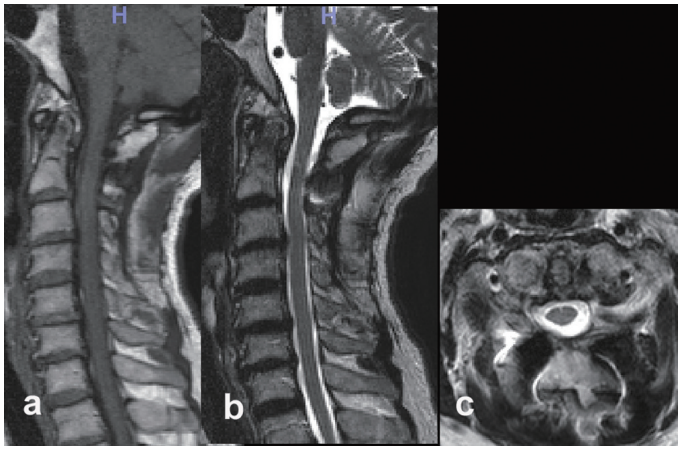

Fig. 3 Postoperative MRI (a: T1-weighted image, b: T2-weighted sagittal image, c: T2-weighted axial image) showing resolution of the pseudotumor and decompression of the spinal cord.

retroodontoid mass had disappeared completely (Fig. 3).

The patient and his family were informed that data concerning the case would be submitted for publication, and they consented.

DISCUSSION

An atlantoaxial subluxation associated with a nontumorous lesion in the posterior region of the odontoid process has been described as a retroodontoid pseudotumor. The initial report by Sze *et al*. indicated that this pathological condition may lead to cervical myelopathy [2]. Retroodontoid pseudotumors have been found frequently in patients with severe rheumatoid arthritis [3, 4]. However, reports of the pathological findings to be the minimal inflammatory reaction and spontaneous resolution after fixation of unstable segment suggest that the soft tissue hypertrophy is caused from a reaction to mechanical stress more than from an inflammatory proliferative process [4, 9, 10]. In the current case, the patient had an 18-year history of torticollis. Computed tomography revealed the atlantoaxial rotatory fixation, accompanied by osteoarthritis of the atlantoaxial joints. This patient also had a 7-year history of senile tremor, which was diagnosed by an experienced neurologist. Frequent rotation of the patient's head due to his tremor was thought to have caused friction of the lateral atlantoaxial facet and the accompanying osteoarthritis. This long-term, rare mechanical stress most likely induced hypertrophic changes of the tissue around the lateral facets. Although the pseudotumor itself was not directly resected, the occipitoaxial stabilization resulted in complete disappearance of the pseudotumor after three years. This suggests that the soft tissue hypertrophy was caused by mechanical stress. This spontaneous disappearance after the occipitoaxial arthrodesis strongly suggests the frequent rotation of the head by the senile tremor as the cause of the pseudotumor.

In selecting the surgical procedure, several methods were considered. If the pseudotumor were caused only by mechanical stress, then atlantoaxial fusion without decompression would have been appropriate. However, if the neurological deficit is progressive and severe, then decompression should be selected because it could take years for the pseudotumor to disappear. In general, atlantoaxial fusion is difficult to perform in decompression surgery because decompression is achieved by resection of the posterior arch of the atlas, which requires a bone graft. Moreover, passage of a sublaminar wire for stabilization of the grafted bone carries the risk of neurological comorbidity when there is limited working space, as was the case here. Thus, we chose the occipitoaxial fusion because the patient's neurological deficit was severe and MRI showed marked canal stenosis.

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