

## Combined Atlantoaxial Rotatory Fixation and Unilateral Counter Occipitoatlantal Subluxation after Minor Trauma

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Rotatory fixation of the atlantoaxial joint is relatively rare in adults but somewhat more common in children. Most of them are reduced spontaneously or after traction. However, combined rotatory injury of atlantoaxial joint and unilateral occipitoatlantal subluxation is a rare disease, and it is often difficult to diagnose accurately. Because the craniovertebral junction is complex and patients with combined atlantoaxial rotatory fixation (AARF) and occipitoatlantal subluxation has a less rotated head than patients with pure AARF. And injury of the occipitoatlantal and atlantoaxial joint can often result upper cervical instability, so early diagnosis is very important. We present a case of the combined AARF and unilateral counter occipitoatlantal subluxation after minor trauma. (Korean J Neurotrauma 2013;9:27-29)

**KEY WORDS:** Atlantoaxial rotatory fixation · Occipitoatlantal dislocation · Arthrodesis.

### Introduction

Rotatory deformities of the atlantoaxial joint may be caused by trauma, upper respiratory tract infection, juvenile rheumatoid arthritis, congenital condition (Down's syndrome, Marfan's syndrome). Although most of them are reduced spontaneously or with traction, in rare cases, they may persist. To describe such persistent rotation, Wortzman and Dewar<sup>7)</sup> in 1968 used the term, rotary fixation of the atlantoaxial joint. In 1977 Fielding and Hawkins<sup>5)</sup> used the term, atlantoaxial rotatory fixation (AARF), because the fixation of the atlas on the axis may occur with subluxation or dislocation. Although AARF in children may be a well-documented, combined AARF and occipitoatlantal subluxation is a rare disease.<sup>3)</sup> Patients with combined AARF and occipitoatlantal subluxation has less rotated and tilted head than those with AARF only, because of counter occipitoatlantal subluxation. So it is often difficult to diagnosis accurately. We report a case of combined AARF and unilateral counter occipitoatlantal subluxation after minor trauma.

### Case Report

A nineteen-year-old girl who had been involved in an unrestrained passenger automobile accident was brought to the emergency department. She presented with diffuse neck pain and limitation of neck rotation. Neurologic examination was normal except that her head was slightly rotated to the left and tilted on the right. Computerized tomography (CT) scans of cervical spine revealed the atlantoaxial rotatory subluxation with 26 degrees, the atlas turned to the left and subluxation with 14 degrees on the occipital condyle (Figure 1). The right condyle of occiput was posteriorly dislocated on the lateral mass of the atlas (Figure 2). The results of vertebral CT angiography were normal. Magnetic resonance imaging demonstrated normal transverse ligament.

At first, the patient was treated with incremental cervical halter traction to a total of 7 kg for 2 weeks. But reduction was unsuccessful. And then she was placed into halo traction after induction of a general anesthetic with injection of muscle relaxant for 2 hours. We checked three dimensional CT every 15 minutes. However, fluoroscope guided halo traction did not produce any mobilization. We thought that combined atlantoaxial rotatory fixation and unilateral counter occipitoatlantal subluxation hinder reduction. So the open reduction was performed. On intraoperative field, C1 rotated to counterclockwise and C1-C2 articular capsule was torn. Reduction was accomplished by counter rotation using bone

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forceps. And C1-C2 screw-rod fixation was done (Figure 3). After operation, the patient was maintained in halo-vest fixation for stabilization of occipitoatlantal subluxation for a month and then a Sternal Occipital Mandibular Immobilizer brace for two months. After removal of brace, she had a good clinical appearance and was able to turn the head without severe neck pain. At the 6 month postoperative visit, she had a stable C1-C2 fusion in anatomical position (Figure 4) and remained neurologically intact.

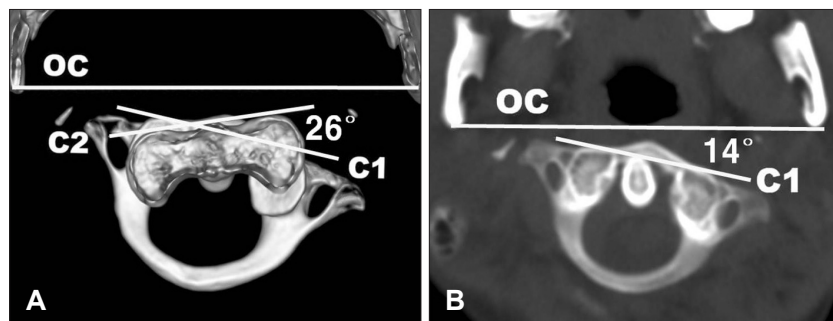
## Discussion

In 1959, Washington<sup>6)</sup> reported a case of a combined atlanto-axial and occipital disorder. In patients with combined atlantoaxial and occipitoatlantal rotatory fixation, the head is gently tilted and slightly rotated because of atlantoaxial rotatory fixation with compensatory counter occipitoatlantal subluxation. In our case, the symptoms of the patient were just limitation of head rotation and slightly tilted and rotated head. At first, it was misdiagnosed as cervical sprain at emergency department because the patient only presented with axial neck pain without neurologic deficit from minor

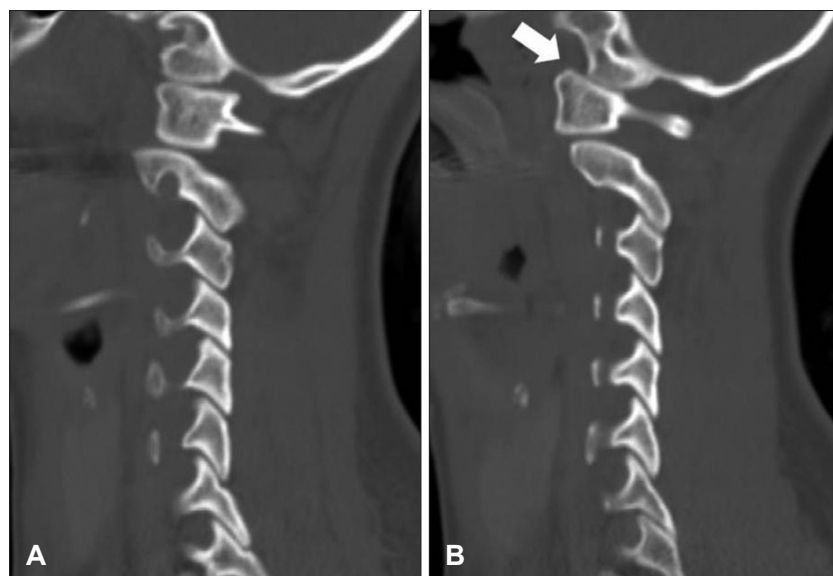
trauma. This pattern of combined dissociation is an uncommon and the exact pathophysiological mechanism of the condition is unknown.<sup>3-6)</sup> But it was probably caused by the violent motion in rotation and traction that caused a compensatory contralateral subluxing of occiput-atlas onto the in-



**FIGURE 3.** Postoperative three-dimensional CT scan (posterior view). Satisfactory reduction has been accomplished and arthrodesis at C1-C2 has been performed in which allogenic bone chips.



**FIGURE 1.** A: Three-dimensional CT scans showing the atlantoaxial rotatory subluxation with 26 degrees. B: C1 turned to the left and subluxation with 14 degrees on the occipital condyle. OC: occipital condyle.



**FIGURE 2.** A: Left parasagittal CT reconstruction showing good articular congruence between the condyle and the lateral mass of C1. B: Right parasagittal CT reconstruction showing posterior dislocation of the condyle on the lateral mass of C1 (arrow).



**FIGURE 4.** Postoperative plain X-ray film (lateral view) showing fusion between C1 and C2.

itial atlas-axis subluxation.<sup>3)</sup> Anatomically, the craniovertebral junction is complex and it is often hard to interpret radiographs of this region. So a complete understanding of the anatomy of the craniovertebral junction is essential to the diagnosis and treatment. Three dimensional CT scanning and CT angiography scanning can be used to demonstrate anatomical location, angle of rotation of each vertebra and injury of vertebral arteries. More recently magnetic resonance imaging is suited to confirm the spinal cord compression and to examine the anatomical integrity of alar and transverse ligament.

Treatment for combined atlantoaxial and occipitoatlantal rotatory fixation is based on the principles of stabilization and fusion. In the literature, two types of therapy could be proposed. The one was treated by undergoing occiput-C2 arthrodesis<sup>2,3,5)</sup> and the other one was treated by C1-C2 arthrodesis and external immobilization using halo vest.<sup>1)</sup> Altongy and Fielding<sup>1)</sup> performed C1-C2 arthrodesis and external immobilization and they believed that normal flexion-extension function of the occipitoatlantal joint was worth

preserving. In our case, the right condyle was posteriorly dislocated on the lateral mass of the atlas, but left side was not dislocated, so we performed C1-C2 arthrodesis and external immobilization. However, extended arthrodesis has to be considered in the event of recurrence. But combined atlantoaxial and occipitoatlantal rotatory fixation has rarely been reported, so there is no established guideline for surgical treatment.

## Conclusion

We report a case of the combined atlantoaxial rotatory subluxation and unilateral occipitoatlantal subluxation after minor trauma. It is often difficult to diagnosis accurately and it can be progressively result in craniovertebral junction instability. So early diagnosis and surgical decision is very important for these patients.

■ The authors have no financial conflicts of interest.

## REFERENCES

- 1) Altongy JF, Fielding JW. Combined atlanto-axial and occipito-atlantal rotatory subluxation. A case report. *J Bone Joint Surg Am* 72:923-926, 1990
- 2) Bouillot P, Fuentes S, Dufour H, Manera L, Grisoli F. Imaging features in combined atlantoaxial and occipitoatlantal rotatory subluxation: a rare entity. Case report. *J Neurosurg* 90(2 Suppl):258-260, 1999
- 3) Clark CR, Kathol MH, Walsh T, el-Khoury GY. Atlantoaxial rotatory fixation with compensatory counter occipitoatlantal subluxation. A case report. *Spine (Phila Pa 1976)* 11:1048-1050, 1986
- 4) Crossman JE, David K, Hayward R, Crockard HA. Open reduction of pediatric atlantoaxial rotatory fixation: long-term outcome study with functional measurements. *J Neurosurg* 100(3 Suppl Spine):235-240, 2004
- 5) Fielding JW, Hawkins RJ. Atlanto-axial rotatory fixation. (Fixed rotatory subluxation of the atlanto-axial joint). *J Bone Joint Surg Am* 59:37-44, 1977
- 6) Washington ER. Non-traumatic atlanto-occipital and atlanto-axial dislocation; a case report. *J Bone Joint Surg Am* 41-A:341-344, 1959
- 7) Wortzman G, Dewar FP. Rotary fixation of the atlantoaxial joint: Rotational atlantoaxial subluxation. *Radiology* 90:479-487, 1968