

Case Report

Atlantoaxial Rotatory Fixation in Adults Patient

Sei Woong Jeon, M.D., Je Hoon Jeong, M.D., Ph.D., Seung Myung Moon, M.D., Sun Kil Choi, M.D.

Department of Neurological Surgery, Hangang Sacred Heart Hospital, College of Medicine, Hallym University, Seoul, Korea

Atlantoaxial rotatory fixation (AARF) in adult is a rare disorder that occurs followed by a trauma. The patients were presented with painful torticollis and a typical 'cock robin' position of the head. The clinical diagnosis is generally difficult and often made in the late stage. In some cases, an irreducible or chronic fixation develops. We reported a case of AARF in adult patient which was treated by immobilization with conservative treatment. A 25-year-old female was presented with a posterior neck pain and limitation of motion of cervical spine after a traffic accident. She had no neurological deficit but suffered from severe defect on the scalp and multiple thoracic compression fractures. Plain radiographs demonstrated torticollis, lateral shift of odontoid process to one side and widening of one side of C1-C2 joint space. Immobilization with a Holter traction were performed and analgesics and muscle relaxants were given. Posterior neck pain and limitation of the cervical spine's motion were resolved. Plain cervical radiographs taken at one month after the injury showed that torticollis disappeared and the dens were in the midline position. The authors reported a case of type I post-traumatic AARF that was successfully treated by immobilization alone.

KEY WORDS : Atlantoaxial subluxation · Adult patient · Radiography · Rotatory fixation · Conservative management · Torticollis.

INTRODUCTION

Atlantoaxial rotatory fixation (AARF) is a rare condition and often under recognized, resulting in an incorrect or delayed diagnosis¹⁷⁾. There is usually a higher incidence in children. The common clinical characteristics are painful torticollis and cock robin position presented with the head tilted to one side and rotated to the other side. Because a clinical diagnosis of AARF is generally difficult, it is often made in the late stage. In some cases, an irreducible or chronic fixation develops¹¹⁾. We report a case of acute post-traumatic AARF in adults that was treated by traction and immobilization.

CASE REPORT

A 25-year-old female was taken to our emergency room after a traffic accident. She complained of severe headache, neck and upper thoracic pain. She suffered from a severe

defect on the scalp with underlying skull exposure and multiple thoracic spine compression fractures, but had no neurological deficit. The patient was admitted to our intensive care unit. After 5 days of conservative management, initial symptoms were improved. However, she complained the neck pain with limitation in range of motion. We decided to review and further skeletal evaluation of the skull base and neck. Cervical spine radiography demonstrated straightening and mild scoliotic change of the cervical spine. An open mouth radiography showed widening of the right C1-C2 joint space with the left C1 lateral mass which is closer to the odontoid process than right one (Fig. 1A). Three-dimensional surface reconstructions of CT (General Electric Light Speed Scanner, Munich, Germany) images revealed the laterally displaced odontoid process to the left C1 lateral mass and a normal atlantodental interval and C1 malrotation to the right, that was defined as AARF (Fig. 1C). However, MRI showed no transverse and alar ligament injury (Fig. 1B). Immobilization with a Philadelphia brace and bed rest were followed. After a few hours, the patient was able to turn normally and rotate her neck again. Thereafter, neck pain was resolved with a rapid improvement in the motion of the cervical spine. The open mouth radiography revealed that the dens were in the midline position (Fig. 1D). She was

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• Address for reprints : Je Hoon Jeong, M.D., Ph.D.
Department of Neurological Surgery, Hangang Sacred Heart Hospital,
College of Medicine, Hallym University, 94-195 Yeongdeungpo-dong,
Yeongdeungpo-gu, Seoul 150-030, Korea
Tel : +82-2-2639-5650, Fax : +82-2-2676-7020
E-mail : neur71@gmail.com

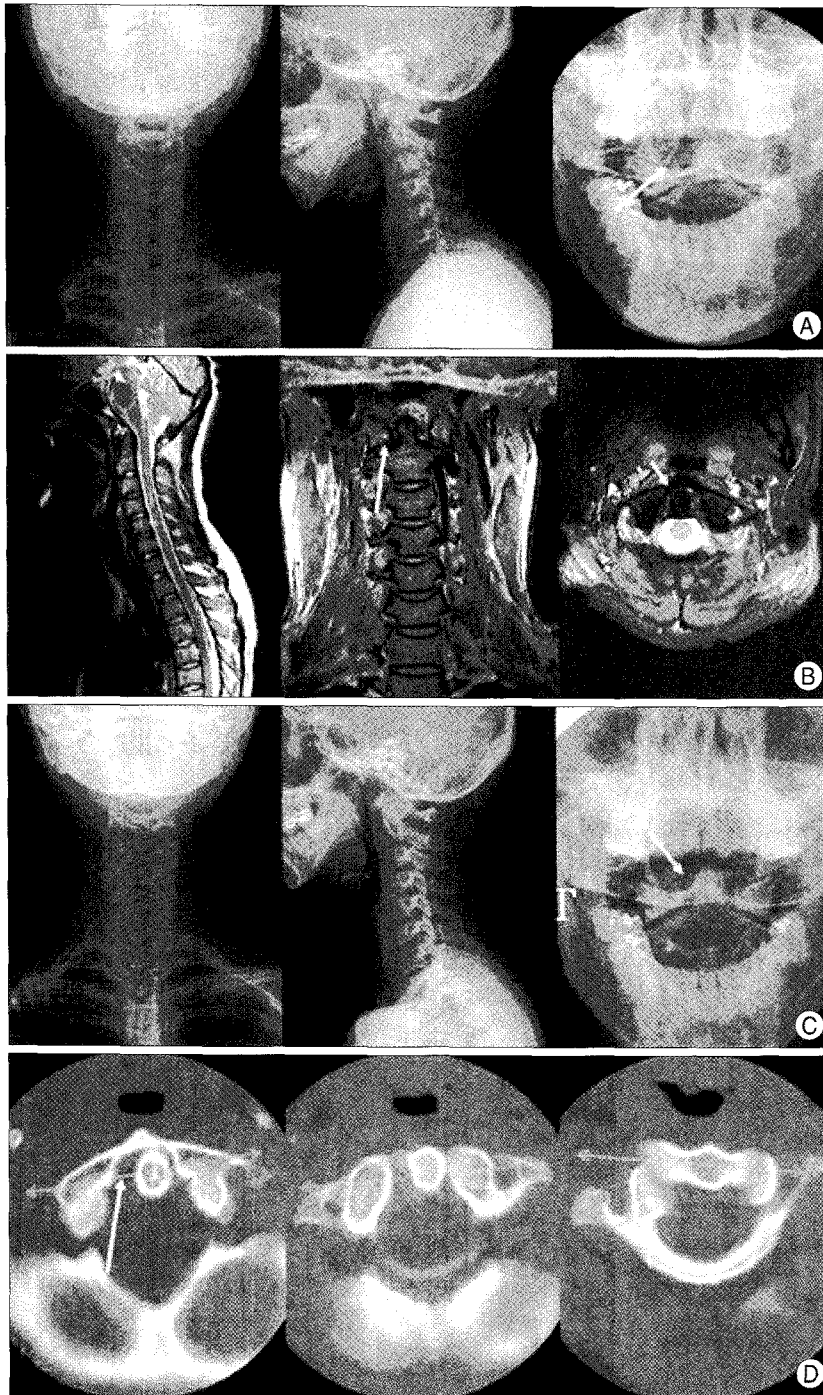


Fig. 1. Radiographs of cervical spine demonstrate mild torticollis presenting as a cock robin appearance and widening of left atlantoaxial joint space with left C1 lateral mass process than right, but no transverse atlantal ligament injury (A, B and C). Computed tomography scan shows right side rotation of atlas with no anterior displacement (C). Post-reduction plain radiograph shows disappearance of scoliosis of cervical spine and displacement of odontoid process (D).

advised to wear a soft cervical collar for another six weeks. At the six months follow-up, she was free from the neck pain and resumed normal activities. These follow-up radiograph images confirmed the atlas and axis has been completely repositioned.

DISCUSSION

Rotatory deformity of the atlantoaxial joint was termed by Wortzman and Dewar in 1968. However, Fielding and Hawkins preferred the term atlanto-axial rotatory fixation (AARF), because the fixation of the atlas on the axis may occur still within the normal range of rotation. The common cause of AARF is generally an infection or a trauma³⁾, but other conditions such as ankylosing spondylitis⁹⁾, metastatic tumor¹⁸⁾, generalized ligamentous laxity^{11,19)}, and eosinophilic granuloma¹¹⁾ have also been reported. The phenomenon of AARF with or without subluxation predominates in children and female regardless of cause^{5,7)}.

Although the pathophysiology of AARF is not well defined, they noticed that the lateral joint capsule was very wide and the transverse and alar ligaments had increased laxity. However, the fixation phenomenon could not be reproduced experimentally¹⁶⁾. The higher incidence of AARF in children is probably attributable to a combination of factors : a large head and underdeveloped neck musculature, a rotational angle greater than 45°, the horizontal configuration of the C1-C2 articular facets, and an increased elasticity of the joint capsules¹²⁾. Therefore, the pediatric population is predisposed to AARF and a differential diagnosis should be made when dealing with acute torticollis in children.

On open mouth radiography, anteriorly rotated C1 lateral mass appears wider and closer to the midline than the lateral mass on the opposite side, and the C1-C2 joint spaces appear asymmetrically. Cervical radiography followed by CT scanning is considered the best method to detect this abnormality²⁰⁾, and three dimensional CT reconstruction is an additional aid in demonstrating subluxation. CT scan also allows an excellent demonstration of abnormal C1-C2 relationships, visualizing the dislocation, determining whether it is unilate-

ral or bilateral, and looking for fractures^{1,2,6,13}. Three-dimensional reconstruction images give a global view of the cervical deformity¹. Magnetic resonance imaging (MRI) can offer direct visualization of tear or avulsion of the transverse ligament⁴. Incidence of AARF in adults patients is lower than that of children¹², and because of skull superimposition on the upper cervical spine, plain radiographs are sometimes difficult to diagnose AARF, especially in emergency state. Even in adult patients, physician needs to be extremely careful about management of patients with limitation of neck motion (LOM) and multiple trauma, especially the head trauma.

The treatment options include immobilization with conservative care, traction, manual reduction, and surgery. Acute traumatic AARF is often reduced easily. Treatments advocated in many cases of less than a 1-month history and minor trauma¹² are immobilization with or without traction and the patients achieve good long term stability. However, the time interval between injury and reduction appears to correlate with rates of recurrence and failure of reduction by non-surgical techniques. Chronic AARF over three months may become irreducible^{8,15}. In 1977, delay in diagnosis averaged 1 year⁵; almost every patient was treated by arthrodesis after several days or weeks of skull traction. Therefore, treatment by cervical traction started as early as possible may cure the patient and can be reduced to a minimum of 24 hr and operative C1-C2 fixation can be prevented. A surgical approach is needed for cases of AARF with spinal instability, neurological involvement, or failure to maintain reduction by conservative measures^{5,10,14}. In our case, posterior neck pain and limitation of motion of neck were resolved spontaneously during traction and bed rest with the neck immobilization by Philadelphia collar allowed full recovery. At 6 months follow-up, she had no posterior neck pain and LOM of neck. Furthermore, she had no neurological deficit with normal atlantoaxial articulation on open-mouth radiograph.

CONCLUSION

The diagnosis of AARF, especially in adult patient with multiple traumas, is easily missed out because the incidence is low and a neurological function is normal. Even in adult patients, AARF should be kept in mind in patient who present with the neck pain and the limitation of neck motion combined with a head trauma. A precise diagnosis and an early treatment are the keys to prevent the operative treatment.

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