Atlanto-Occipital Rotatory Dislocation: A Case Report and Systematic Review

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Key words
- Atlanto-occipital dislocation
- Atlanto-occipital dissociation
- Atlanto-occipital rotatory dislocation
- Occipitoatlantal dislocation

Abbreviations and Acronyms
AOD: Atlanto-occipital dislocation
AOJ: Atlanto-occipital joint
AORD: Atlanto-occipital rotatory dislocation
CCI: Condylar—C1 interval
CT: Computed tomography
MRI: Magnetic resonance imaging
TAARS: Traumatic atlantoaxial rotatory subluxation/dislocation

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■ BACKGROUND: **Atlanto-occipital rotatory dislocation (AORD) has rarely been reported in the literature; for this reason, the clinicoradiologic characteristics of this injury are not well described.**

■ METHODS: **We describe the case of a 67-year-old man who sustained a cervical spine trauma. He reported only neck pain and was neurologically intact. A computed tomography scan showed a rotatory displacement of the atlanto-occipital joints associated with a widened condylar—C1 interval; in addition, magnetic resonance imaging showed injuries to the stabilizing ligaments of this area. A systematic literature review was also performed to identify previous cases of patients with AORD.**

■ RESULTS: **The patient was treated with craniocervical fixation from occipital to C1, achieving a good outcome. The literature review yielded 9 cases of patients with AORD. Compared with patients with atlanto-occipital dislocation, patients with rotatory dislocations have a less severe degree of displacement of the atlanto-occipital joints and better clinical outcome.**

■ CONCLUSIONS: **Compared with previously classified atlanto-occipital dislocations, AORD is an independent and unique variation. AORD presents with different biomechanical, clinicoradiologic, and prognostic characteristics and represents an important addition to the spectrum of atlanto-occipital dislocation injuries.**

INTRODUCTION

Atlanto-occipital dislocations (AODs) occur predominately as ligamentous injuries and are usually the result of high-energy trauma such as motor vehicle accidents or falls from heights, involving large acceleration and deceleration forces.[1] Traditionally, AOD has been classified into 3 types[2]: type I involves an anterior displacement of the occiput from the atlas; type II is a longitudinal distraction with separation of the same structures; and type III denotes posterior displacement of the occiput. Different patterns of injury such as rotatory or lateral displacement are not included in this classification. This classification method is based on the direction of displacement and the anatomic position of the atlanto-occipital joints (AOJs); however, other characteristics such as stability or severity of injury are not considered. For these reasons, the classification loses some of its clinical usefulness in the management of these patients, and as result, different classification methods have been suggested.[3,4]

Atlanto-occipital rotatory dislocation (AORD) is an uncommon injury, which has been reported in the literature mainly as case reports.[1,5-10] Because of the rarity of this entity, a detailed description of the characteristics of this condition is missing. We present the case of a patient with a traumatic AORD and a comprehensive review of literature and analysis of this uncommon type of spinal injury.

METHODS

Study Selection

The case of a patient who experienced an AORD while swimming in the ocean is reported. In addition, a systematic literature review was also performed. A comprehensive literature search of PubMed and Scopus was performed in accordance with PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines. Because we consider that AORD is an under-reported condition, cases of AORD were intentionally sought in articles reporting patients with AOD. Further, the references for all search-selected articles were also reviewed for potential cases.

The search of publications was performed using the following terms: "atlantooccipital rotatory dislocation," "atlantooccipital dislocation," "occipitoatlantal dislocation," "atlantooccipital dissociation," and "craniocervical dislocation." The search was extended to all available English language articles from 1980 to May 2017. The selected starting date of search was 1980 because computed tomography (CT) has been used more widely since that date.

Cases were included in the review only if patients had documented AORD on imaging and if the articles had sufficient information including radiologic details,

clinical status, type of treatment, and outcome. Exclusion criteria were animal studies, cadaveric or forensic studies, literature reviews, studies reporting radiologic analysis, description of surgical techniques, cases of atraumatic AOD, letters or correspondence, studies reporting only outcome, articles reporting different diseases from AOD, and articles with incomplete clinical information or those with inadequate imaging. Titles and abstracts were initially reviewed to identify articles with positive exclusion criteria.

Data Extraction

Data were extracted from eligible cases. Specific information was obtained from eligible articles. Collected data included 1) age, 2) type of accident, 3) clinical status, 4) associated spine injuries, 5) radiologic findings, 6) treatment, and 7) outcome.

RESULTS

Case Description

History. A 67-year-old man had an accident while he was swimming in the ocean. He was caught and rolled over by a wave and was driven into the water, hitting his head on the seabed. He experienced transient numbness and weakness in the 4 limbs, which completely improved in the following minutes. In the emergency room, the patient reported only neck pain; on examination, bruises were observed in the left frontotemporal area and he was noted to have a normal motor and sensory function of the upper and lower limbs.

Radiologic Tests. Imaging tests showed an unusual pattern of AOD characterized by rotatory displacement of the AOJs; in addition, other traumatic injuries were observed in the same area. Conventional radiographs showed only degenerative spine changes. The CT scan showed the following findings (**Figure 1A–F**): 1) the traditional radiographic techniques for diagnosis of AOD were normal on a midsagittal view; 2) on parasagittal views, a bilateral posterior displacement of the occipital condyles was observed predominately on the right side; moreover, the mean distance of the condylar–C1 interval (CCI) was increased predominately on the left side (right, 2.5 mm; left, 4.3 mm); 3) on coronal view, the left AOJ was distracted but almost preserved the alignment with C1 lateral mass while the right occipital condyle was laterally displaced; in addition, a comminuted fracture was also visualized between the basion and the upper border of the atlas and a type III condylar fracture was observed in the right occipital condyle. In a different coronal cut through the dens, the lateral dens–atlas space was increased on the right side; 4) on axial view, a rotatory AOD was observed, in which the occipital condyles were rotated and displaced to the right in a posterolateral direction. A CT three-dimensional reconstruction provided a different view of these injuries (**Figure 2A–C**). Sagittal magnetic resonance imaging (MRI) (**Figure 3A–C**) showed an intact tectorial membrane; on a coronal view, the atlanto-occipital and atlantoaxial articular capsules were observed with increase in the intensity and distraction injury and on axial view, the right alar ligament was observed attached to the bone fragment of the condylar fracture.

Treatment and Outcome. Because the patient was a tourist, he was transferred to his country of origin to continue treatment. He was treated with posterior occipitocervical fixation. He was contacted 4 months later; at that time, he did not report any neurologic symptoms.

Systematic Review

The initial literature search yielded 688 articles. After removal of duplicates, the title and abstract of 494 articles were screened and based on exclusion criteria, 341 articles were eliminated. After this initial filter, 153 articles were assessed for eligibility, of which 146 were excluded because of lack of evidence indicating AORD. Only studies showing patients with radiographic evidence of AORD were selected for the final review. Thus, a total of 7 studies encompassing 9 patients were eligible for analysis (**Figure 4**). **Table 1** shows the characteristics of these patients who were identified from previous publications[1,5-10] and also includes our patient, totaling 10 patients with AORD.

This review shows that AORD may occur at any age. The age range observed in these cases was 9–67 years. The mechanism of injury varied by patient, with several being low energy.

Dickman et al.[7] reported a series of 14 patients with AOD. Ten of these patients died acutely and 1 more patient died 3 months after the initial injury. The remaining 3 patients achieved long-term survival and had a significant neurologic recovery. All these patients had an identifiable AORD. Among these 3 patients, the initial plain radiographs did not show findings consistent with AOD, and surgical findings showed instability and a high degree of hypermobility of the craniovertebral junction. All developed good osseous fusions. One of these cases is of special interest: a child who was initially comatosed and whose MRI showed an epidural hematoma compressing the brainstem. After an operation to remove the hematoma and stabilization, the patient showed great neurologic improvement, which suggests that the clinical status was secondary to the epidural hematoma and not to the rotatory dislocation.

Banna et al.[6] described a patient with a unilateral AOD. However, the analysis of the imaging tests showed that this injury was an AORD. Anatomically, it is not possible to observe a pure unilateral displacement of the AOJ because both occipital condyles are displaced at the same time and therefore, the lateral condylar displacement must be bilateral. In addition, the rotational injury was observed in the axial CT scan. This patient was neurologically intact and achieved a good outcome after treatment with bracing. There are several studies reporting patients with lateral AOD[11-13]; however, they had incomplete imaging and imaging findings from review of some of the available radiographs were suggestive of a rotational displacement.

Lee et al.[10] reported on 1 patient who experienced an AORD. She was treated conservatively and achieved a functional outcome, albeit she remained with torticollis 7 months later. Angiography was performed, showing unilateral injury to the vertebral artery. More than 50% of angiograms of patients with craniocervical injuries may show a vascular injury, in either the vertebral or the carotid arteries.[14]

The remaining patients showed a good neurologic status[1,5,8,9] and achieved a satisfactory outcome.