Case Report

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Surgical Management of Adult Traumatic Atlantoaxial Rotatory Subluxation with Unilateral Locked Facet; Case Report and Literature Review

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ABSTRACT

Atlantoaxial rotatory subluxation (AARS) is rarely occurred in adults with trauma as the most common cause. In type A and B it is usually managed with close reduction and external brace; however, in nonresponsive cases, surgical interventions might be needed. Our patient is a 21-year-old man with neck pain and torticollis after a car turn- over. There was C1-C2 rotatory subluxation with left side locked facet and C1 rotation about 40 degrees relative to C2 on computed tomography without evident of ligamentous injury in magnetic resonance imaging (MRI). However, during the first 48 hours, two tries of close reduction using Gardner cervical traction under fluoroscopy were failed. Thus, the patient underwent open reduction of the subluxation and atlantoaxial fixation (Harm's technique) with subsequent relief of pain and torticollis. This a rare case of traumatic AARS type A with unilateral locked facet joint in an adult patient which needed surgical manipulation for reduction. The management of the AARS in adults should be individualized in each patient.

Keywords: Atlantoaxial rotatory subluxation (AARS); Atlantoaxial fixation; Trauma; Adult.

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Introduction

The atlantoaxial facet joint is the primary pivotal point and fulcrum of movements in the region. The odontoid process and the atlantodental joint assist in keeping the movements under a check or control [1]. The nature of atlantoaxial dislocation can be gauged by visualization of the facets [2, 3]. The adult atlantoaxial rotatory subluxation (AARS) is a rare condition in craniovertebral junction with

only few cases reported in the English literature [4-8]. The condition is mainly associated with trauma and the diagnosis is usually a dilemma to the spine surgeons [9]. The delay in diagnosis and management might result in neurological deficit or respiratory suppression and death due to upper cervical cord injury [10, 11]. The treatment of this condition is controversial and both conservative and surgical treatment have been described previously [12-14]. The rarity of the AARS in adults, has limited our experience in both diagnosis and management of the condition. Thus, we herein report an adult case of AARS which was successfully managed surgically with open reduction and atlantoaxial fixation.

Case Report

A 21-year-old man was referred to our center due to car accident from a primary center with Philadelphia collar. The patient was a victim of a car turnover accident following a car-car collision while driving in road with speed higher than 120 km/hr. He was restrained by the seat-belt. He reported a severe acceleration-deceleration movement of neck with no history of direct trauma to head and neck. He was conscious with no neurological deficit; however, he complained of neck pain and limitation of range of motion (ROM). On inspection, his neck was fixed in position while lateral bended to the left side and rotated to the right side. Computed tomography (CT) of the craniovertebral junction revealed AARS with left side locked facet joint and C1 rotation about 40 degrees relative to C2 (Figure 1). Magnetic resonance imaging (MRI) revealed mild injury of transverse ligament with no evidence of obvious rupture. The patient was diagnosed to suffer from traumatic anterior atlantoaxial facet (or type A) subluxation. Conservative management with cervical traction was decided for the patient. Cervical traction with Gardner was applied without appropriate response up to 5kg along with appropriate administration of muscle relaxants, sedatives and painkillers. Thus, we decided to reduce the AARS through surgical approach. The patient underwent atlantoaxial fixation with bilateral C1 lateral mass screws fixation bilateral C2 pedicular screws (Harm's technique). Reduction of the deformity was performed after C2 ganglion sectioning and gentle manipulation of the locked facet joint. The construct was fixed in compression to obtain normal upper cervical lordosis (Figure 2). After operation, the neck deformity and loss of motion was recovered completely with resolved neck pain. Cervical CT-scan confirmed the proper reduction of AARS (Figure 3). The patient had an uneventful hospital course and was discharged 3 days postoperatively. In 6-month follow-up there was no cervical pain and the patient was neurologically intact. However, the patient had 30-degree rotation limitation to each side. But there was no flexion and extension limitation of motion.

Discussion

AARS is usually encountered in pediatric patients especially in the presence of underlying diseases with ligamentous laxity [7, 15]. However, trauma-induced AARS in adult patients is a very rare entity with quite few reports in the English literature [4-6, 11]. In 1970s, Fielding and Hawkins [16], described 4 different types for AARS according to the

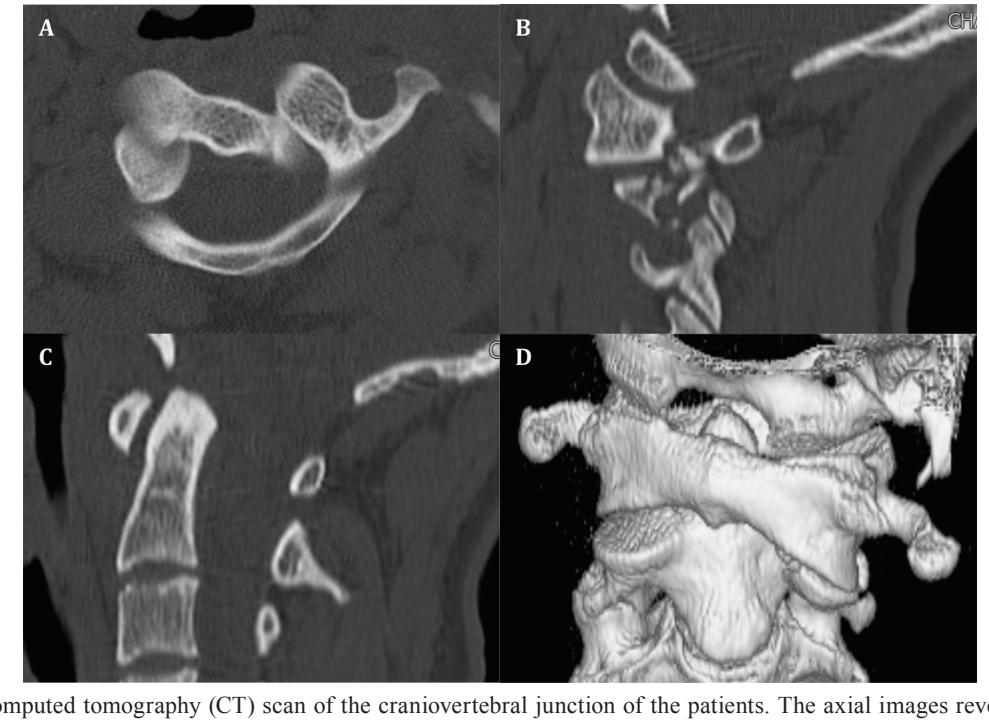

Fig. 1. The computed tomography (CT) scan of the craniovertebral junction of the patients. The axial images reveal atlantoaxial rotatory subluxation (AARS) with left facet locked **(A)**; the sagittal images through the left C1-C2 facet joint **(B)** and mid-sagittal views **(C)** clearly demonstrate the AARS with left locked facet. The coronal 3D reconstruction images reveal rotation of the C1 about 40 degrees relative to C2 **(D).**

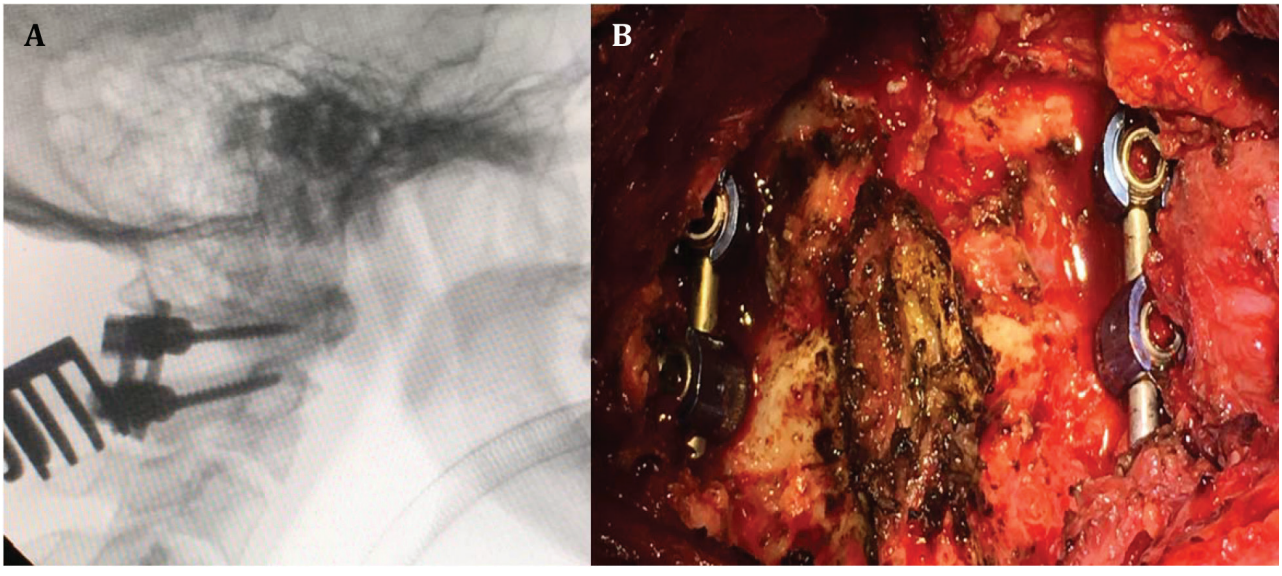

Fig. 2. The intraoperative lateral fluoroscopy of the atlantoaxial region demonstrating complete reduction of the atlantoaxial rotatory subluxation (AARS) with C1-C2 fixation **(A)**. The intraoperative posterior view of the surgical site demonstrating posterior fixation and reduction of the AARS **(B)**.

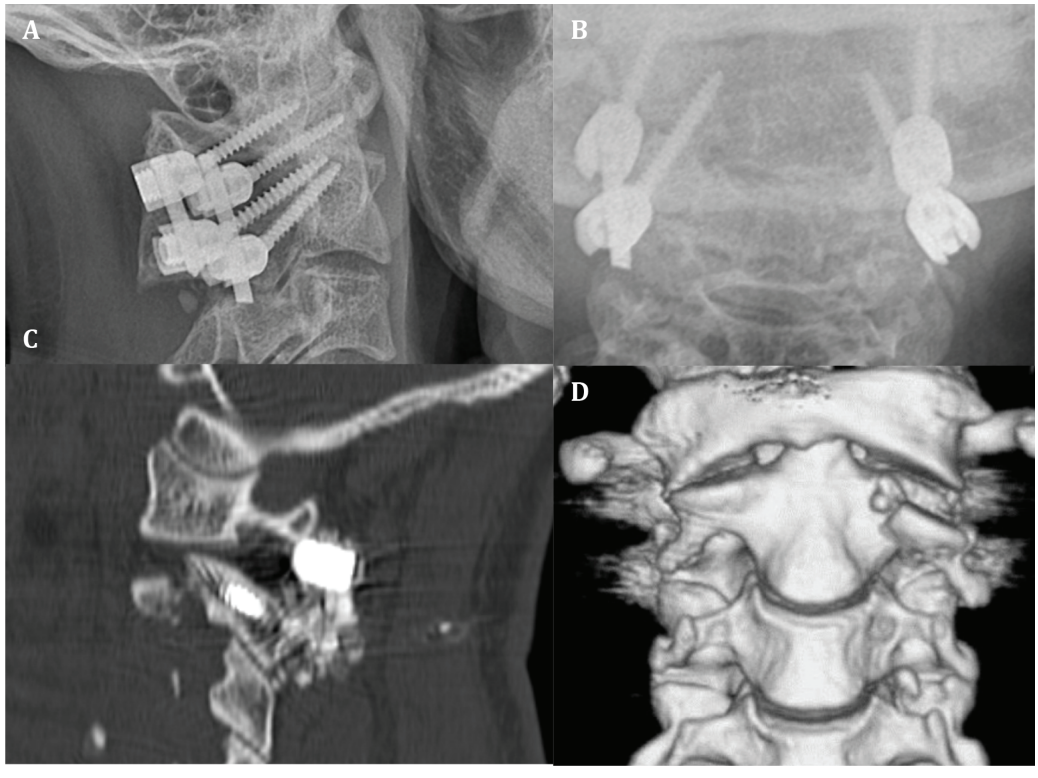

Fig. 3. The 6-month postoperative radiography of atlantoaxial spine in lateral **(A)** and anteroposterior **(B)** views demonstrating complete reduction of the atlantoaxial rotatory subluxation (AARS), fusion of the atlantoaxial joint and appropriate instrumentation. The sagittal computed tomography (CT)-scan of the atlantoaxial joint through left facet demonstrating complete reduction **(C)**. The postoperative coronal 3D reconstruction CT-scan of the patient revealing complete reduction of the AARS.

severity of the injury. Type A which is the mildest form is referred to translation of facet joints within the normal range of atlantoaxial rotation with no increase in atlantodental interval (ADI). If transverse ligament is ruptured, atlantoaxial rotation of more than normal range with increased ADI may occur which defined as AARS type B. With more severe injuries, bilateral forward or backward dislocation of atlantoaxial joint and subsequent spinal canal stenosis will be expected (type C and D) [16]. The most common type among reported cases in both pediatric and adult population is type A and other types are much rarer and require more complicated management [7].

Nonetheless, regarding the etiology of intense trauma in most adult cases of traumatic AARS likelihood of more severe or fatal types, although still rare, are higher as compared to pediatric cases [7, 9].

The typical clinical presentation includes cervical pain with torticollis and contralateral neck bending, known as Cock-Robin position [8, 12, 14]. Although some atypical accompanying clinical feature has also been reported [5]. Prompt diagnosis should be made on the basis of clinical presentations combined with appropriate imaging modalities such as CT-scan and MRI. As the condition is missed easily and the diagnosis is usually made based on the clinical suspicion. The neck limitation of motion, pain and abnormal head position in patients with multiple trauma and high speed injuries is the key to diagnose the condition with appropriate imaging techniques. Unfortunately, currently there is no available guideline for diagnosis of these conditions. Cervical CT-scan is usually used to evaluate bony structures and classify AARS. The side of subluxation and the degree of rotation will be best depicted in coronal view of CT-scan [3]. MRI will be most helpful in evaluation of ligamentous structures especially in cases with C1-C2 instability [10, 17].

While management of AARS is generally aimed to decrease neck pain; however, the restoration of cervical alignment and prevention of neurological deficit is the main goal. The choice of best therapeutic approach is still controversial due to rarity of disease and it is better to be individualized in each case [7, 9]. Therapeutic approaches include closed reduction and external immobilization with Halo-Vest or Philadelphia collar for 6-12 weeks or surgical reduction and fixation. The time of diagnosis is also so critical in treatment planning since early management can increase the success rate of closed reduction, reducing the need for further surgical interventions [16].

Our case was diagnosed properly in first hours after trauma with type A severity and no neurological deficit, leading to early management with 4-5 kg traction. However, due to lack of expected response after 48 hours, he was scheduled for surgical posterior C1-C2 fixation. While closed reduction of atlantoaxial joint is generally enough in most early diagnosed AARS cases of type A and type B [9, 10], our case was resistant to closed reduction even after 2 tries. This can be partly explained by the presence of locked facet joint in left side, requiring more careful rotatory manipulation during traction. This case further emphasizes on the individualized management of AARS while even in the AARS type A surgical intervention should be kept in mind especially in cases with simultaneous locked facet joint.

In conclusion, traumatic AARS in adults is rare entity and should be diagnosed early to prevent further neurological injuries. The management is controversial and should be individualized. In type A, conservative management with cervical traction followed by immobilization is recommended. However, in unresponsive cases, surgical intervention is required to restore the cervical alignment and reduce the pain and instability.

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Conflicts of Interest: None declared.

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