CASE REPORT

Surgical Management of Adult Traumatic Atlantoaxial Rotatory Fixation with Hangman Fracture: Case Report and Literature Review

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Abstract

Atlantoaxial rotatory fixation (AARF) is a rare type of adult cervical spine injury. The classic symptoms are painful torticollis and limited neck range of motion. To avoid catastrophic consequences, early diagnosis is necessary. The present study presents the successful treatment of a scarce case of adult AARF with a Hangman fracture and a comprehensive literature review. A 25-year-old man presented to the trauma bay with left-sided torticollis after a motor vehicle accident. Cervical computed tomography scans revealed type I AARF. Torticollis resolved after cervical traction with partial reduction, and posterior C1-C2 fusion was performed as part of the therapy. Recognition of AARF after trauma requires a high index of suspicion, and early diagnosis is critical for the achievement of the best possible patient outcomes. Since the combination of a Hangman fracture and C1-C2 rotatory fixation is complex and unique, it must be tailored to the associated injuries.

Level of evidence: IV

Keywords: Adult, Atlantoaxial rotatory fixation (AARF), Axis fracture, Hangman fracture, Torticollis

Introduction

The atlantoaxial joint is one of the most active joints in the cervical spine, accounting for more than 60% of the 90° rotation to either side. Several craniocervical ligaments, particularly the transverse and alar ligaments, help pivot C1 around C2 dens. Atlantoaxial rotatory subluxation or fixation (AARF) is relatively uncommon, especially in adults, and can lead to devastating neurological outcomes in case of a delayed diagnosis.

Torticollis, defined as a "cock-robin" posture of the neck, is the most prevalent manifestation of AARF followed by suboccipital headache[1]. Since the condition is easily missed and is usually diagnosed based on clinical suspicion, neck motion restriction, pain, and abnormal head position in patients with multiple trauma and high-energy injuries are the keys to raising suspicion of AARF and requesting additional imaging, such as Computed thermography (CT) scan and magnetic resonance imaging (MRI).

A timely diagnosis is critical in treatment planning since early management can increase the success rate of closed reduction and reduce the need for further surgical interventions[2,3]. Less than 100 cases of adult C1-C2 rotatory fixation have been reported in the last century, and relatively little is known about the clinical manifestations and management of the disease. This report presents a unique combination of a Hangman fracture associated with type II (Fielding and Hawkins classification) AARF.

Case Presentation

Following a collision with a lamppost, a 25-year-old man with no notable medical history was referred to our tertiary trauma center. According to the primary trauma survey, the patient had an occipital headache, severe cervical pain, and limited neck range of motion. The pain in the cervical region was more severe on the right side of the neck, radiating from the back of the neck into the skull. The physical examination revealed that he had stable vital signs, a Glasgow Coma Scale

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score of 15, and light-reactive pupils. Furthermore, the patient denied having any symptoms of focal neurological impairment, such as changes in vision or hearing, speech, focal weakness, numbness, or tingling.

The radiologic assessment included CT scans of the cervical spine without contrast and neck MRI. The CT scans revealed an asymmetry of the odontoid lateral mass interspaces, as well as posterior displacement of C1 lateral mass on C2 on the left and anterior displacement on the right, 3 mm atlanto-dental interval, bilateral pars fracture with less than 3 mm of subluxation of C2 on C3 with no angulation, and prevertebral soft tissue edema from C3 to C5 [Figure 1]. The cervical MRI revealed C1-C2 malalignment, prevertebral soft tissue edema, and C1/C2 joint effusion [Figure 1]. Moreover, there was no evidence of ligament damage. The brain MRI results indicated no abnormalities in the posterior fossa. The rotatory displacement occurred without anterior displacement of C1 on C2, and the transverse ligament was intact; hence, Fielding and Hawkins classification of type I AARF is applicable.

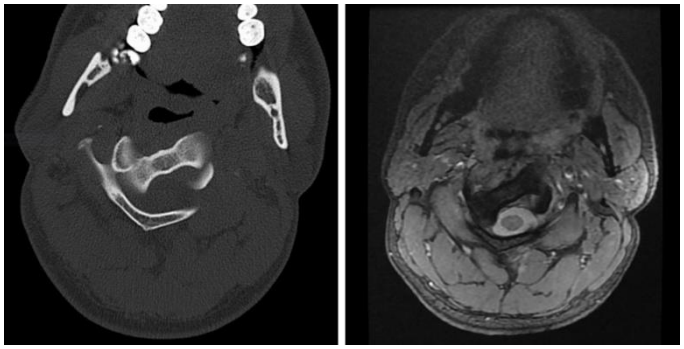

Figure 1. (Left) Axial computed tomography and (Right) Axial magnetic resonance imaging display bilateral rotatory deformity/dislocation of the axis on a normally positioned atlas

The patient received emergency cervical traction with Gardner-Wells skull tongs (5 kg for 48 h). However, the rotatory subluxation did not improve despite the application of cervical traction, and also torticollis persisted. Intraoperative traction, open reduction with gentle manipulation, and posterior C1/C3 fusion were used to release the facet and resolve the torticollis [Figure 2]. After an uneventful postoperative course, he was discharged with a rigid neck brace.

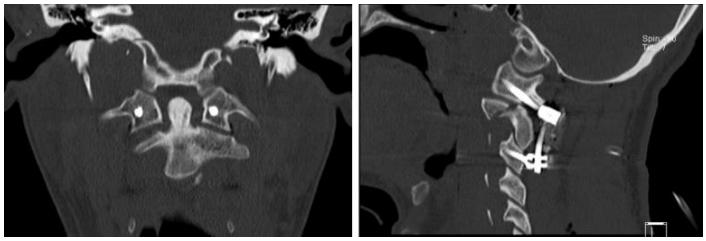

Figure 2. (Left) Coronal and (Right) sagittal computed tomography scan revealed the postoperative construct with C1-C3 fixation and restoration of alignment

Discussion

The AARF in combination with traumatic C2-spondylolisthesis is extremely rare. A comprehensive PubMed search (English literature from 1907 to 2022) resulted in 16 cases of AARF with axis fractures, while only one was a Hangman fracture [Table 1]. The reported patients had an average age of 46.1±18.7. Most cases were male (n=12, 75%), and the most common mechanism of trauma was a fall (n=9, 56.3%) followed by motor vehicle accidents (n=5, 31.3%), bicycle rollover (n=1), and unspecified trauma (n=1). The lateral mass (n=10, 62.5%) and the odontoid process (n=9, 56.2%) were the most commonly fractured parts of the axis. Anderson D'Alonzo type II odontoid fractures (n=6) were twice as common as type III (n=3).

The present study reports a scarce case of AARF with hangman fracture that was successfully diagnosed and treated. The first case of a C1–C2 rotatory fixation with hangman fracture was reported by Chaudhary et al. in 2015.[4] The authors described a 26-year-old man who suffered from type IIa Hangman fracture and AARF due to a motor vehicle accident.

In adults, AARF is a rare type of cervical spine injury, and its rarity often results in missed or delayed diagnoses. A high index of suspicion and the use of diagnostic imaging appropriately will allow for an early diagnosis and prompt treatment. Painful torticollis and limited neck range of motion are the cardinal symptoms of AARF, followed by sub-occipital headache caused by greater occipital nerve stretching or C2 root compression.[5]

Rarely, posterior fossa symptoms, such as vertigo and ataxia, have been described, most likely as a result of vertebral artery compression.[3, 6] the craniocervical junction should also be assessed with a CT scan as part of the radiologic assessment. An MRI is essential to determine the integrity of the ligamentous structures, including the transverse and alar ligaments and the spinal cord.[4, 5]

In this case, two classification systems are relevant, namely the Levine and Edwards classification and the Fielding and Hawkins classification [Table 2]. Traumatic C2-spondylolisthesis is classified using the Levine and Edwards system.[7] In the 1970s, Fielding and Hawkins classified AARF into four types based on the severity of the damage.[8] Types I and II account for most documented cases in pediatric and adult populations, whereas other types are less common and require more complex management [Table 1]. In the case of this study, the patient had a Type I Hangman fracture (bilateral pars fracture with <3 mm of subluxation of C2 on C3 and no angulation) and a C1-C2 rotatory dislocation.

The combination of traumatic C2-spondylolisthesis and AARF poses a unique challenge. The final decision requires consideration of the type and instability of the fracture, as well as the rotatory dislocation. For most stable hangman fractures, nonsurgical management with rigid immobilization has been widely accepted.[9] Likewise, nonsurgical treatment, early cervical traction, and anatomic reduction followed by 6-12 weeks of immobilization have successfully treated AARF.[10]However, C1-C2 arthrodesis is recommended for unstable fractures, neurologic deficits, or those that do not respond to skeletal traction and conservative management.[11]

Table 1. Summary of reported adult cases of atlantoaxial rotatory fixation with associated axis fractures

Author/Year	Age/Gender	Trauma	F&H Class	Associated Injuries	Management
Gahlot, 2020	47, M	Fall	Type III	C2 body, odontoid (type III), lateral mass, and pedicle	Posterior C1-C2 fixation
Meyer et al., 2019	61, F	Fall	Type I	C2 lateral mass, apical and alar ligaments, and facet capsule	Posterior C1-C2 fixation
	77, M	Fall	Type II	Odontoid (type II), apical and alar ligaments, and facet capsule	Posterior C1-C2 fixation
	79, F	Fall	Type I	Odontoid (type II), C1, apical and alar ligaments, and facet capsule	Occipito-cervico-thoracal fusion and decompression
Chang et al., 2018	63, M	Fall	Type II	Odontoid (type III) and C2 lateral mass (bilateral)	Halo traction, manual derotation, and halo vest (12 weeks)
Opoku-Darko et al., 2018	20, F	MVA	Type I	Odontoid (type II)	Halo traction, Manual derotation, and posterior C1-C2 fixation
Chaudhary et al., 2015	26, M	MVA	Type II	Hangman (type IIa), alar ligament, and facet capsule	Halo traction and posterior C1-C3 fixation
Bellil et al., 2014	56, F	MVA	Type I	C2 lateral mass	Awake manipulation and halo vest (12 weeks)
Yang et al., 2010	37, M	Fall	Type I	Odontoid (type III) and C2 lateral mass	Skull traction and halo vest (12 weeks)
Spoor et al., 2008	43, M	Bicycle roll-over	N/A	C2 body, odontoid (type II), and lateral mass	Halo traction and halo vest (16 weeks)
Kim et al., 2007	34, M	Fall	Type I	C2 lateral mass, and transverse ligament (partial)	Manual reduction under G/A and posterior C1-C2 fixation (Harms technique)
Fuentes et al, 2001	24, M	Fall	Type IV	Odontoid (type II)	Manual reduction under G/A, anterior Odontoid fixation, and posterior C1-C2 fixation
Chen et al., 2001	29, M	MVA	Type II	C2 body, lamina, and C1 lateral mass	Manual reduction under G/A, and halo vest (12 weeks)
Cheng et al., 2000	43, M	Fall	N/A	C2 body, odontoid (type II), and lateral mass (comminuted)	Closed reduction and posterior C1-C2 fixation
Moore et al., 1995	65, M	MVA	Type I	C2 lateral mass and transverse process	Gardner-Well tongs traction and posterior C1-C2 fixation
Kowalski et al, 1987	34, M	Trauma	Type I	C2 lateral mass	Halo traction

MVA: motor vehicle accident
F & H Class: Fieldings and Hawkins classification of AARF

Table 2. Most common relevant classifications

Fielding and Hawkins classification of atlantoaxial rotatory fixation:

- **Type I**: Rotatory dislocation without anterior displacement, intact transverse ligament, and the dens acting as the pivot point
- **Type II**: Rotatory dislocation with anterior displacement between 3-5 mm, injured transverse ligament, and the opposite facet acting as the pivot point
- **Type III**: Anterior rotatory dislocation of both facet joints with anterior displacement > 5 mm and injured transverse ligament and facet capsules
- **Type IV**: Posterior rotatory dislocation of both facet joints

Levine and Edwards classification of Hangman fractures:

- Type **I**: Minimal fracture displacement with <3 mm anteroposterior translation, no angulation, and intact C2/3 disc (stable injury)
- **Type II**: Fracture displacement with >3 mm anteroposterior translation, angulation of 10 degrees or less, vertical fracture line, and disrupted C2/3 disc and posterior longitudinal ligament (unstable injury)
- **Type IIa**: Type II fracture with minimal or no translation but significant angular deviation, angulation >10 degrees, and horizontal/oblique fracture line (unstable injury)
- **Type III**: Type II fracture with bilateral facet joint dislocation

Given an unstable type IIa Hangman fracture, Chaudhary et al. avoided cervical traction and proceeded with posterior fusion.

Various surgical techniques exist to address such complex injury patterns. Both anterior and posterior procedures have been described. For anterior cervical discectomy and fusion, Smith-Robinson, transoral, or extraoral approaches have been introduced.[4, 12] anterior approaches may help treat traumatic C2-C3 spondylolisthesis with disc herniation and medullary compression. Still, they are more complicated and can injure the facial nerve, hypoglossal nerve, superior laryngeal nerves, and carotid sheath.

The posterior approach has a high fusion rate of 87-100% and a lower complication rate[4]. For combination injuries that cause atlantoaxial instability, the posterior approach offers flexibility (including C1 fixation) and is superior to anterior cervical plating from a biomechanical point of view.

Several factors determine the surgical technique for a posterior C1-C2 rotatory fixation, including the stability of the injury site, the integrity of the transverse ligament, and the posterior C1 arch. Nonunion risk is higher with C1-C2 wiring or posterior fixation with hooks. On the other hand, these procedures require that the posterior C1 arch remains intact. A unilateral or bilateral transarticular C1-C2 fixation can be performed to reduce the risk of nonunion. However, this technique requires a complete reduction of the rotatory dislocation prior to screw fixation. In addition, transarticular screw fixation is associated with an increased vascular risk, which is especially problematic if there is vertebral dissection. The Goel-Harms technique can reduce rotation and translation between C1 and C2, even if the posterior C1 arch is damaged. To avoid an intraoperative vascular lesion, the trajectory of vertebral arteries in C2 must be carefully identified pre-operatively.

The patient in this report sustained a complex injury with type I Hangman fracture and Type I AARF without any neurological deficits. Initially, cervical traction was applied. However, the dislocation failed to respond, and open reduction with posterior fusion was planned. Although most type I and type II rotatory fixation patients with an early diagnosis can often benefit from closed reduction and conservative management, closed reduction was ineffective in this case. This was primarily attributable to a locked facet joint on the left, which required more careful rotatory manipulation during traction.

The surgical fixation resulted in solid fusion, avoiding delayed union that can occur with nonoperative treatment. Therefore, the present case report emphasizes the value of individualized AARF management and demonstrates that in cases with persistent locked facet joints or associated atlas or axis fractures, surgical intervention should be considered for both types I and II C1-C2 rotatory fixation.

Conclusion

For cases of AARF type I and II diagnosed early, closed reduction and immobilization with a halo ring or cervical collar are standard treatment options. When conservative treatment fails, as is common in chronic irreducible cases or cases with associated axis fractures and instability, surgical intervention is required to restore cervical alignment and relieve pain.1[3, 14]

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