

Traumatic atlantoaxial rotatory fixation in an adult patient

María A. García-Pallero¹ · Cristina V. Torres¹ · Juan Delgado-Fernández¹ · R. G. Sola¹

Received: 11 August 2016 / Revised: 24 November 2016 / Accepted: 5 December 2016
© Springer-Verlag Berlin Heidelberg 2016

Abstract



Introduction Atlantoaxial rotational fixation (AARF) is a rare entity in adults, with only a few cases reported in the English literature and often associated with a traumatic mechanism. It is an underdiagnosed condition that must be taken into account in the initial assessment of all cranio-cervical trauma. Both diagnostic and therapeutic delay may be a potential cause of severe neurological damage or even death of the patient. The therapeutic management is controversial given the difficulty of achieving optimum stability and permanent reduction.

Methods and results A 28-year-old woman was involved in a traffic accident a week before coming to the emergency with rotation and irreducible cervical flexion from

trauma and severe neck pain. CT and MRI column were performed and showed a cervical spinal AARF with transverse and alar ligaments intact and preserved atlantoaxial distance (Fielding I). The patient was treated by progressive cervical traction with 5 kg and manual reduction was completed in 24 h. Subsequently, an external immobilization was performed by cervical rigid collar for 16 weeks. The clinical course was good, with the patient regaining full mobility with cervical neck pain improvement.

Conclusions The purpose of this paper is to show a case of a young woman with a posttraumatic AARF successfully treated conservatively. This case delineates the difficulties in diagnosing this pathology, as well as the challenges encountered in its management.

Keywords Atlantoaxial joint · C1–C2 fixation · Cervical spine · Rotatory dislocation · Traction

Case presentation

A 28-year-old woman was involved in a traffic accident; she was driving a car when she suffered a frontal collision and the car turned right over in mid-air. She did not lose consciousness and left the car by her own. She went to the hospital; they performed a cervico-dorsal X-ray and she was discharged with diagnosis of whiplash trauma. 6 days later, she decided to go to the hospital for persistent and severe neck pain and paresthesias in the first three fingers of her left hand. A physical examination revealed irreducible cervical rotation to the right side and flexion to the left side since the trauma.

She underwent a CT scan of the cervical spine where a rotational subluxation between C1 and C2 without fracture

✉ María A. García-Pallero
magpallero@gmail.com

¹ Department of Neurosurgery, University Hospital La Princesa, C/Diego de León 62, 28006 Madrid, Spain

lines was objectified. The cervical MRI confirmed the AARF with right C1–C2 lateral recess enlargement, occupied by an increase of soft tissue and the integrity of the transverse and alar ligaments. The atlantoaxial distance was preserved and there were no signs of spinal cord compression.

Diagnostic imaging section

See Figs. 1, 2, 3.

Historical review of the condition

The unilateral rotational subluxation of the atlantoaxial joint is a relatively common pathology in cervical trauma [1]. However, the rotational bilateral subluxation is a rare entity, particularly in the adult population. There are only a few cases reported in the English literature, of which only 16 are of Fielding type I and no other associated lesions (Table 1).

In children, it is a common condition, usually caused by a mild trauma or, in many cases, by a respiratory infection of the upper respiratory tract (Grisel syndrome) [2–4]. The



Fig. 1 3D reconstruction of cervical CT showing an AARF of C1 on C2

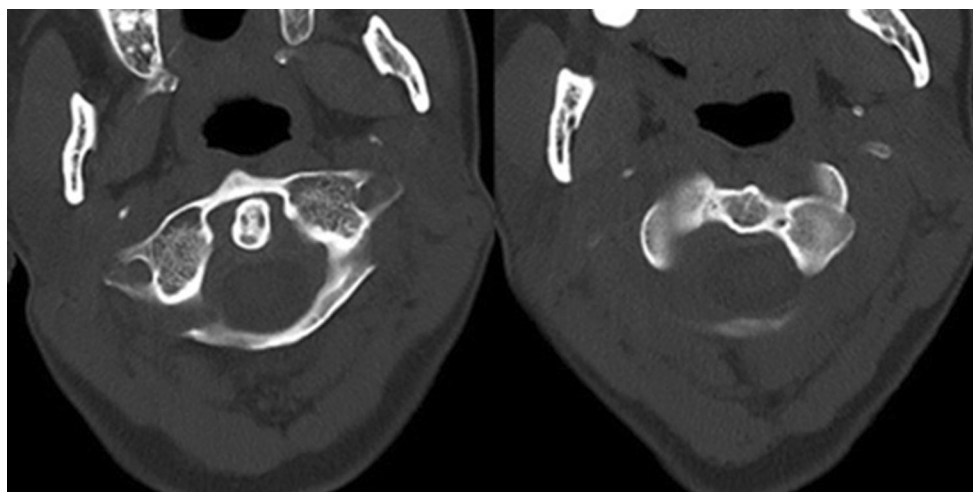


Fig. 2 Cervical axial CT image. Head in a neutral position showing dens asymmetry relative to lateral masses of axis and rotary displacement without anterior displacement (Fielding type I)

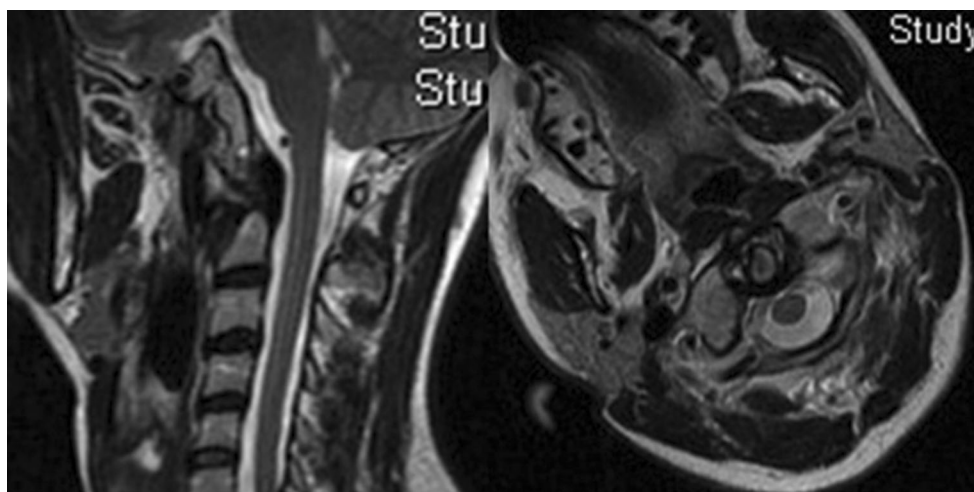


Fig. 3 T2-weighted magnetic resonance image showing intact alar and transverse ligaments

Table 1 Previously described cases of traumatic atlantoaxial rotatory fixation Fielding 1, in the adult population, in the English literature

Author	Sex and age (years)	Mechanism	Time to diagnosis (days)	Management	Results
Jones [13]	Male, 18	Sports accident	0	Halter traction + cervical collar	No sequelae
Robertson [14]	Male, 18	Sports accident	0	Halter traction + SOMI brace	30° loss of rotation to right
Wise [15]	Female, 29	Motor vehicle accident	0	Gardner-Well tongs traction + halo vest	No sequelae
Castel [16]	Male, 41	Sports accident	30	Cervical traction + Minerva jacket	No sequelae
Sinigaglia [17]	Female, 26	Automobile accident	45	Cervical traction + halo vest	Neck stiffness and headache
	Female, 21	Automobile accident	1	Cervical traction + halo vest	No sequelae
	Male, 29	Automobile accident	1	Cervical traction + cervical collar	No sequelae
Wang [18]	Female, 44	Undescribed	180	Halo vest	Weakness in both hands
Singh [12]	Female, 25	Automobile accident	0	Cervical traction + halo vest	No sequelae
Jeon [7]	Female, 25	Automobile accident	5	Cervical traction + cervical collar	No sequelae
Marti [19]	Female, 24	Stretching and cervical rotation by herself	1	Cervical traction + halo vest	No sequelae
Venkatesan [3]	Female, 20	Automobile accident	0	Cervical traction + cervical collar	Occipital pain
	Female, 52	Automobile accident	0	Cervical traction + cervical collar	Loss of 10° of cervical rotation and occipital pain
Maida [20]	Female, 27	Automobile accident	Few days	Manual reduction + cervical collar + posterior fusion	Loss of 30° of cervical rotation and occipital neuralgia
Meza Escobar [21]	Female, 19	Automobile accident	0	Cervical traction + cervical collar	No sequelae
Min Han [5]	Male, 22	Automobile accident	0	Cervical traction + cervical collar	No sequelae

reason that it predominates in children is due to the higher relative proportion of the size of the head relative to the body; the insufficient development of the neck muscles; the elasticity of the articular capsule and the large angle of rotation between C1 and C2; and the horizontal configuration of the facet joints between atlas and axis [5–7].

In adults, the mechanism of injury is high-energy trauma such as car accidents, falls or sports accidents [2]. Given the nature of the trauma, patients often have other complications such as damage to the articular cartilage or fractures in the facet joints [1]. They may also have associated spinal cord injury, which will depend on the integrity of the transverse ligament, the displacement of C1 on C2 and the invasion of the spinal canal [8].

The pathophysiology of the AARF is not well defined. The atlantoaxial joint primarily contributes to about 50% of axial neck rotation and the transverse and alar ligaments provide the majority of biomechanical ligamentous stability. The transverse ligament and facet joint capsule prevent anterior translation of C1 on C2. The alar ligaments connect the posterolateral apex of the odontoid with the lateral aspect of the foramen magnum bilaterally, mainly limiting anterior shift of the atlas on the odontoid and excessive rotation of C1 on C2 to 50° [3, 9]. There is evidence from cadaveric dissection and magnetic resonance imaging studies to suggest that the pathologic mechanism by which rotatory subluxation occurs is through disruption of the facet capsule followed by alar ligament disruption. The lateral mass of the atlas rotating posteriorly locks behind the ipsilateral lateral mass in severe forms of atlantoaxial rotatory instability [3, 10].

Fielding and Hawkins described four types of AARF [11]. This classification, which has been widely accepted, correlates with increased risk of spinal instability and potential neurologic impairment. The type I indicates a displacement of the facet joints without a corresponding increase in atlantodens interval, and rotation is within the normal range of normal atlantoaxial rotation; this is the only case that can happen without rupturing ligaments and where the dens acts as a pivot. The transverse ligament may rupture, producing an increase in atlantodens interval by 3–5 mm (Type II). If the atlas is dislocated bilaterally with displacement more than 5 mm, this would be a type III, and if it does back narrowing the spinal canal, it would be a type IV [11].

The clinical findings show the typical position of painful torticollis with lateral neck flexion and contralateral rotation position known as Cock-Robin [11, 12]. A high index of suspicion, physical examination and appropriate imaging tests are the bases in obtaining an early diagnosis of this disease.

The CT cervical study is considered of choice to detect this type of injury, since it gives us an excellent

demonstration of abnormal relations between C1 and C2, allows us to determine whether it is unilateral or bilateral, and we can also see if there are any associated fractures. It also allows us a three-dimensional reconstruction of images that give us an overview of the cervical deformity [9, 11]. The MRI is essential to assess the spinal cord, to see if there is a disc fragment in the canal and to see the integrity of the ligamentous structures, especially alar and transverse ligaments [8].

Rationale for treatment and evidence-based literature

The therapeutic management is controversial because of the rarity of the disease, and generally requires some form of immobilization, either internal or external.

The goals of treatment are to restore the range of pain-free movement, prevent or reverse any neurological compromise and restore spinal stability [9]. The options include conservative treatment with immobilization, traction and manual reduction, or surgery [4, 6]. Also, an early diagnosis is important to treat early and, thus, reduce the probability of permanent neurological damage [3, 6].

There are some authors like Weisskopf et al. [10] who have reported surgical treatment by posterior fixation, but many other authors [9, 11, 12] recommend conservative treatment using manual cervical traction and external immobilization, either a halo or rigid collar for 6–12 weeks; this achieves a good rotational stability in most cases; in these cases, surgical treatment would be reserved for spinal instability, neurological deficits, when there is a delay in diagnosis or when conservative treatment failed [1].

In our case, the patient was treated by progressive cervical traction and manual reduction. An external immobilization was then performed by cervical rigid collar for 16 weeks, achieving good stability in the imaging of successive control. Conservative management was selected due to the spinal stability.

Procedure

The patient was taken to the operating room and was treated by progressive cervical traction up to 5 kg and fluoroscopically guided manual reduction, which was completed in 24 h. She underwent a cervical CT scan at 72 h, which showed the reduction of AARF with discreet rotating component. She was discharged after 7 days with neck pain improvement and disappearance of paresthesias. Subsequently, an external immobilization was performed

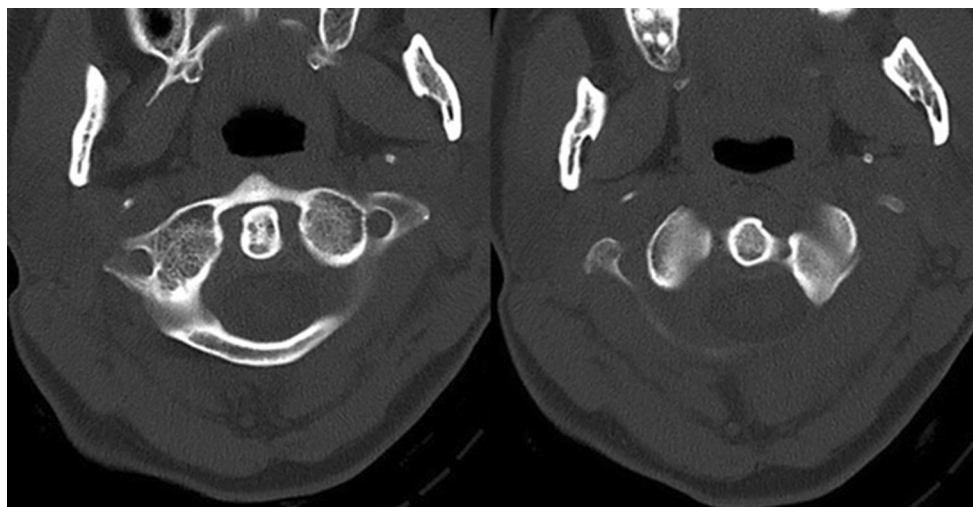


Fig. 4 Cervical axial CT image post reduction showing the restoration of anatomic alignment

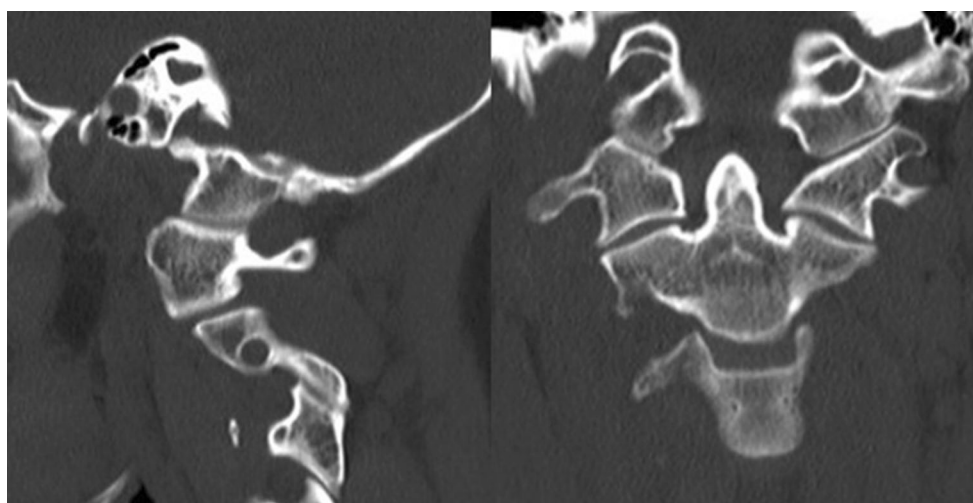


Fig. 5 Cervical sagittal and coronal CT image post reduction showing optimal stability

by cervical rigid collar for 16 weeks. Neck pain persisted for several months, but with rehabilitation and medical treatment, her condition improved.

Procedure imaging section

See Figs. 4 and 5.

Outcome

After 4 years of follow-up, the clinical outcome is excellent. She has recovered cervical mobility and has only slight neck pain when performing intense exercise.

Compliance with ethical standards

Conflict of interest None of the authors has any conflict of interest to disclose.

References

1. Moore KR, Frank EH (1995) Traumatic atlantoaxial rotatory subluxation and dislocation. *Spine (Phila Pa 1976)* 20:1928–1930
2. Crook TB, Eynon CA (2005) Traumatic atlantoaxial rotatory subluxation. *Emerg Med J* 22:671–672. doi:[10.1136/emj.2003.013045](https://doi.org/10.1136/emj.2003.013045)
3. Venkatesan M, Bhatt R, Newey ML (2012) Traumatic atlantoaxial rotatory subluxation (TAARS) in adults: a report of two cases and literature review. *Injury* 43:1212–1215. doi:[10.1016/j.injury.2012.01.013](https://doi.org/10.1016/j.injury.2012.01.013)
4. Ishii K, Toyama Y, Nakamura M, Chiba K, Matsumoto M (2012) Management of chronic atlantoaxial rotatory fixation. *Spine*

- (Phila Pa 1976) 37:E278–E285. doi:[10.1097/BRS.0b013e31823cc2ea](https://doi.org/10.1097/BRS.0b013e31823cc2ea)
5. Min Han Z, Nagao N, Sakakibara T, Akeda K, Matsubara T, Sudo A, Kasai Y (2014) Adult traumatic atlantoaxial rotatory fixation: a case report. *Case Rep Orthop* 2014:593621. doi:[10.1155/2014/593621](https://doi.org/10.1155/2014/593621)
 6. Weisskopf M, Naeve D, Ruf M, Harms J, Jeszenszky D (2005) Therapeutic options and results following fixed atlantoaxial rotatory dislocations. *Eur Spine J* 14:61–68. doi:[10.1007/s00586-004-0772-7](https://doi.org/10.1007/s00586-004-0772-7)
 7. Jeon SW, Jeong JH, Moon SM, Choi SK (2009) Atlantoaxial rotatory fixation in adults patient. *J Korean Neurosurg Soc* 45:246–248. doi:[10.3340/jkns.2009.45.4.246](https://doi.org/10.3340/jkns.2009.45.4.246)
 8. Chaudhary SB, Martinez M, Shah NP, Vives MJ (2015) Traumatic atlantoaxial dislocation with Hangman fracture. *Spine J* 15:e15–e18. doi:[10.1016/j.spinee.2014.12.150](https://doi.org/10.1016/j.spinee.2014.12.150)
 9. Bellil M, Hadhri K, Sridi M, Kooli M (2014) Traumatic atlantoaxial rotatory fixation associated with C2 articular facet fracture in adult patient: case report. *J Craniovertebr Junction Spine* 5:163–166. doi:[10.4103/0974-8237.147083](https://doi.org/10.4103/0974-8237.147083)
 10. Willauschus WG, Kladny B, Beyer WF, Gluckert K, Arnold H, Scheithauer R (1995) Lesions of the alar ligaments. In vivo and in vitro studies with magnetic resonance imaging. *Spine (Phila Pa 1976)* 20:2493–2498
 11. Fielding JW, Hawkins RJ (1977) Atlanto-axial rotatory fixation. (Fixed rotatory subluxation of the atlanto-axial joint). *J Bone Joint Surg Am* 59:37–44
 12. Singh VK, Singh PK, Balakrishnan SK, Leitao J (2009) Traumatic bilateral atlantoaxial rotatory subluxation mimicking as torticollis in an adult female. *J Clin Neurosci* 16:721–722. doi:[10.1016/j.jocn.2008.07.082](https://doi.org/10.1016/j.jocn.2008.07.082)
 13. Jones RN (1984) Rotatory dislocation of both atlanto-axial joints. *J Bone Joint Surg Br* 66:6–7
 14. Robertson PA, Swan HA (1992) Traumatic bilateral rotatory facet dislocation of the atlas on the axis. *Spine (Phila Pa 1976)* 17:1252–1254
 15. Wise JJ, Cheney R, Fischgrund J (1997) Traumatic bilateral rotatory dislocation of the atlanto-axial joints: a case report and review of the literature. *J Spinal Disord* 10:451–453
 16. Castel E, Benazet JP, Samaha C, Charlot N, Morin O, Saillant G (2001) Delayed closed reduction of rotatory atlantoaxial dislocation in an adult. *Eur Spine J* 10:449–453
 17. Sinigaglia R, Bundy A, Monterumici DA (2008) Traumatic atlantoaxial rotatory dislocation in adults. *Chir Narzadow Ruchu Ortop Pol* 73:149–154
 18. Wang YF, Mu-Huo Teng M, Sun YC, Yuan WH, Chang CY (2008) Torticollis due to atlantoaxial rotatory fixation. *J Clin Neurosci* 15:316–318. doi:[10.1016/j.jocn.2006.10.023](https://doi.org/10.1016/j.jocn.2006.10.023)
 19. Marti JJ, Zalacain JF, Houry DE, Isakov AP (2011) A 24-year-old woman with neck pain. *Am J Emerg Med* 29:473.e471–473.e472. doi:[10.1016/j.ajem.2010.04.016](https://doi.org/10.1016/j.ajem.2010.04.016)
 20. Maida G, Marcati E, Sarubbo S (2012) Posttraumatic atlantoaxial rotatory dislocation in a healthy adult patient: a case report and review of the literature. *Case Rep Orthop* 2012:183581. doi:[10.1155/2012/183581](https://doi.org/10.1155/2012/183581)
 21. Meza Escobar LE, Osterhoff G, Ossendorf C, Wanner GA, Simmen HP, Werner CM (2012) Traumatic atlantoaxial rotatory subluxation in an adolescent: a case report. *J Med Case Rep* 6:27. doi:[10.1186/1752-1947-6-27](https://doi.org/10.1186/1752-1947-6-27)