Delayed Spontaneous Reduction of Traumatic Pediatric Atlantoaxial Rotatory Subluxation

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Abstract

Pediatric atlantoaxial rotatory subluxation (AARS) is a rare finding with various etiologies and treatment recommendations. Etiologies include both traumatic and nontraumatic causes. The diagnosis is suggested by clinical presentation and confirmed with imaging. Various forms of management have been discussed in the literature. However, no overall consensus has been established.

We present 2 pediatric cases showing delayed spontaneous reduction of traumatic AARS. Images from computed tomography (CT) were used for initial diagnosis and to track management progression. Although the subluxation persisted on the 1-month follow-up CT in Case 1, the final CT images in both cases showed spontaneous reduction with anatomic positioning of C1 and C2.

These cases demonstrate that delayed spontaneous reduction of traumatic pediatric AARS is possible with conservative treatment. Active reduction via traction may not be necessary. In the absence of compelling surgical indications to the contrary, a conservative approach to management of traumatic pediatric AARS is warranted.

Pediatric atlantoaxial rotatory subluxation (AARS) is a rare finding with various etiologies and unclear treatment recommendations. Etiologies of this condition include both traumatic and nontraumatic causes. The diagnosis is suggested by clinical presentation and confirmed with imaging modalities that often include computed tomography (CT).[1] Presenting symptoms include: neck pain, limited range of motion (ROM), and torticollis in the classic "cock robin" position. The differential diagnoses include: atlanto-occipital injuries, lower cervical injuries, congenital cervical spine anomalies, genetic conditions with ligamentous laxity (Marfan syndrome and Down syndrome), autoimmune disease (rheumatoid arthritis), Grisel syndrome (pharyngitis, cervical abcess, adenoiditis, tonsillitis, and otitis media), tumors, and drug-induced torticollis (ie, phenothiazines).[1-7] Trauma should be considered in any case in which a child presents with acquired torticollis. AARS is the most common cause of traumatic torticollis.[2,3] Various forms of management have been discussed in the literature. Successful treatment with cervical collar immobilization, halo-vest immobilization, and surgical stabilization have all been reported; however, no overall consensus has been established. Although many studies use CT for diagnosis and to track progress of various management choices, confirmation of reduction is often lacking.

We present 2 pediatric cases showing delayed spontaneous reduction of traumatic AARS. The patients' guardians provided written informed consent for print and electronic publication of these case reports. Images from CT were used for initial diagnosis and to track management progression. Although the subluxation persisted on the 1-month follow-up CT in Case 1, the final CT images in both cases showed spontaneous reduction with anatomic positioning of C1 and C2.

Case Reports

Case 1

A 14-year-old adolescent girl presented with neck pain following a motor vehicle accident (MVA). Physical examination showed pain and limited ROM in her cervical spine. Although her neurologic examination was limited by a left forearm fracture, she showed no evidence of spinal cord injury. Initial images (**Figures 1A-1D**) obtained by CT and magnetic resonance imaging (MRI) confirmed a C1/C2 subluxation. Asymmetric widening of the left lateral atlanto-dens interval and a small fragment of bone inferomedial to the left occipital condyle were evident. (**Figure 1B**) Transverse ligament of the atlas appeared intact (**Figure 1C**) preventing anterior translation of C1.

Initial management consisted of closed reduction with Gardner-Wells tong traction and placement in a hard cervical collar. The patient was discharged home with careful instructions and a 2-week follow-up. She remained neurologically stable and her neck pain subsided. A 1-month follow-up CT showed residual subluxation (**Figures 2A, 2B**). The patient

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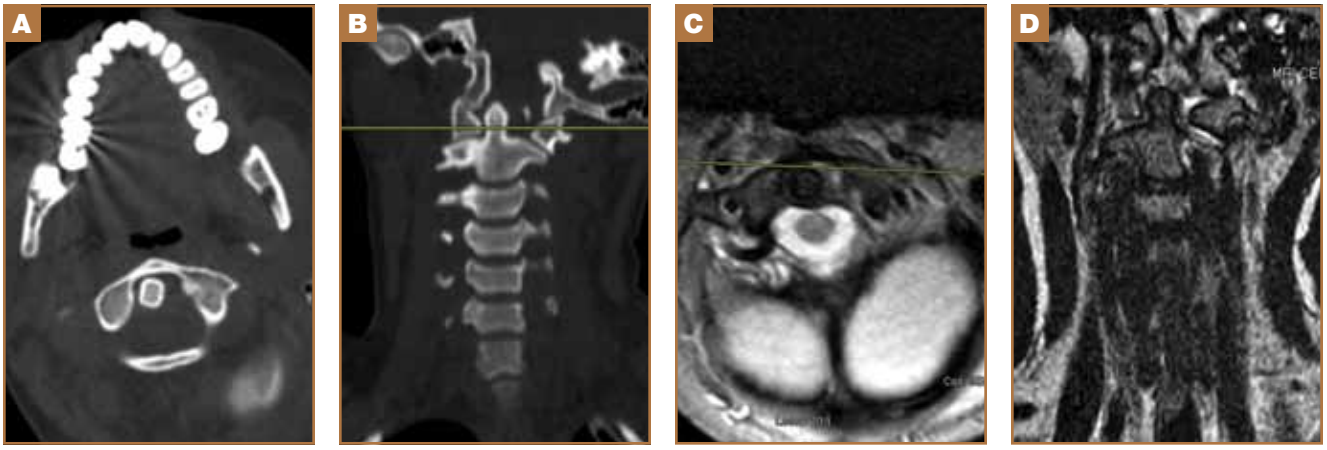

Figure 1. Imaging findings at initial visit. (A) Axial CT scan showing C1/C2 rotatory subluxation. (B) Coronal CT reconstruction showing C1/C2 subluxation and a bone fragment lateral to the left side of the odontoid process with asymmetric widening of the left lateral atlanto-dens interval. (C) Axial MRI without contrast showing intact transverse ligament of the atlas. (D) Coronal MRI showed no identifiable ligamentous injury.

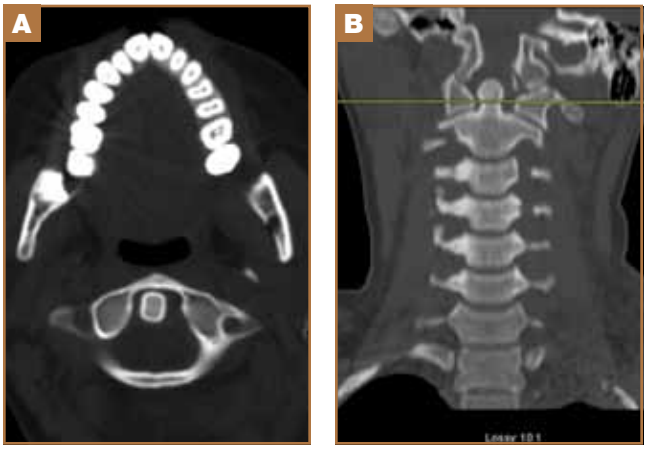

Figure 2. Imaging findings at 1-month follow-up. (A) Axial CT scan showing residual C1/C2 subluxation. (B) Coronal CT reconstruction showing residual C1/C2 subluxation and bone fragment inferomedial to the left occipital condyle.

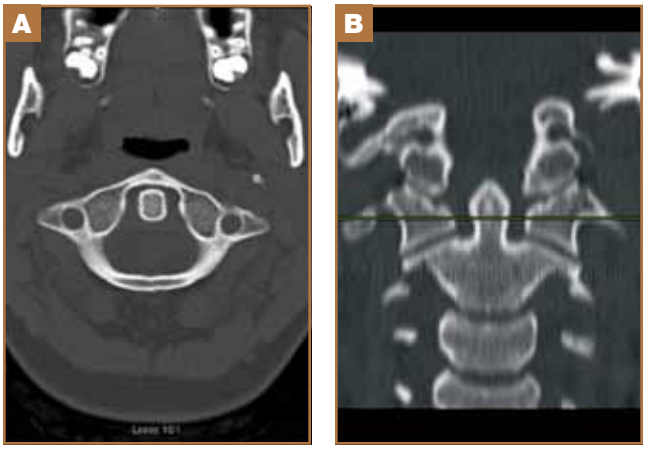

Figure 3. Imaging findings at 6-month follow-up. (A) Axial CT scan showing reduced C1/C2 rotatory subluxation with anatomical alignment. (B) Coronal CT reconstruction showing reduced subluxation of C1/C2 with anatomical alignment.

had minor neck pain with good cervical ROM with no neurological symptoms. At this time, it was decided to discontinue wearing the collar and repeat a cervical CT at her 6-month follow-up. Final CT images (**Figures 3A, 3B**), obtained 6 months post-injury, showed spontaneous reduction with anatomical alignment of C1/C2. The patient demonstrated full, painless, cervical ROM.

Final CT images, obtained 6 months post-injury, showed spontaneous reduction with anatomical alignment of C1/C2. The patient demonstrated full, painless, cervical ROM.

Case 2

A 5-year-old girl presented to the emergency department with neck pain after hitting her head on the end of a swimming pool sliding board. She was neurologically intact. X-ray showed no fractures in the cervical spine. At this time, she was diagnosed with a cervical strain and was sent home on baclofen and ibuprofen.

The patient returned to the pediatrician's office with mild neck pain and difficulty turning her head to the right. CT imaging showed C1/C2 subluxation with asymmetric widening of the right lateral atlanto-dens interval (**Figure 4A**).

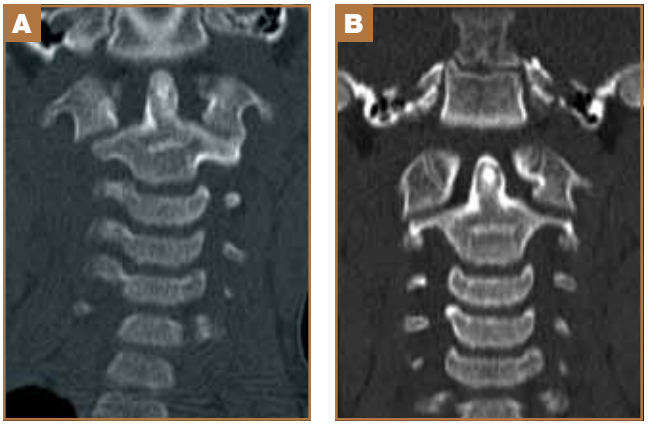

Figure 4. (A) Initial coronal reconstruction CT scan showing C1/C2 subluxation with asymmetric widening of the right lateral atlanto-dens interval. (B) Coronal reconstruction CT scan at 4 months after injury showing reduced subluxation of C1/C2 with anatomical alignment.

Conservative management was continued and follow-up was scheduled. At her 4-month post-injury follow-up, full, painless, cervical ROM was demonstrated and CT imaging showed spontaneous reduction with anatomical alignment of C1/C2 (**Figure 4B**).

Discussion

Rotation is the primary function of the atlantoaxial joint. The primary stabilizer of the joint is the transverse ligament of the atlas which limits excess flexion (excess anterior shift of the atlas on the axis).[2] Alar ligaments are secondary stabilizers that limit excessive rotation and prevent the anterior shift of the atlas on the axis.[2] The pediatric spinal column has unique anatomical and biomechanical features compared to adults. Such unique features include a large head-to-body ratio, which allows greater flexion/extension with bending forces; great elasticity contributing to hypermobility; more horizontal alignment of articulating upper cervical facets; and anterior sliding resulting from the wedge-shaped vertebral bodies.[8,9] These factors predispose children to such conditions as AARS. The transition from a skeletally immature cervical spine to a skeletally mature cervical spine that occurs during the adolescent phase of growth likely represents a unique time period where pediatric and adult injury patterns will be observed. In the presented cases, the 14-year-old demonstrated a bony avulsion that was evident on CT, whereas the 5-year-old demonstrated an apparently pure soft-tissue injury.

The common etiologies of traumatic AARS are MVAs, falls, sports, and recreational injuries.[8,10] Most pediatric patients with traumatic AARS have torticollis in which the head is in the classically described "cock robin" position (ie, lateral flexion to one side with head rotation and slight flexion to the opposite side). Typical presentation in nonsevere trauma includes neck pain with limited ROM and normal neurologic examination as found in the present cases.

Diagnosis is made by careful history, physical examination, and imaging. Radiographs are usually done initially; however, the films may not be relevant due to the position and limited ROM of the head. The normal anatomical position can be obscured in a plain lateral radiograph of the C-spine due to head tilt (classic positioning in AARS). Furthermore, the anterior-posterior position which gives the best view of the odontoid (open mouth view) may not be achievable due to the degree of rotation and/or pain of the patient.[11] Although static CT scans were successfully used in the 2 cases presented, dynamic CT has been recommended in the literature for the diagnosis of AARS.[3] In patients with traumatic AARS, dynamic CT has been suggested to be the superior imaging method when compared to plain radiograph and standard CT because it can show loss of normal C1/C2 rotation when the head is maximally rotated to the right and left.[3,11] The use of CT imaging also provides a strategy for accurately tracking treatment progression as shown in the present case (**Figures 1A-1D, 2A-2B, 3A-3B, 4A-4B**). Because dynamic CT is not a common study and may be unfamiliar to many technicians, the logistics can be difficult. Conventional cervical CT (with pediatric dose reduction) was successfully used in the 2 cases presented and may be a more desirable "first line" test as it avoids the additional radiation from multiple scans during a dynamic test.

Management of traumatic AARS remains controversial although various forms have been suggested in the literature. Traumatic AARS is often easily reduced following a prompt diagnosis however, increasing the duration between injury and treatment has been correlated with rates of failed reduction by conservative methods and reoccurrence.[6] Conservative management consisting of traction and cervical stabilization via rigid neck collar, shown to be effective in several reported cases[7,9,10,12] while others only required simple head-halter traction.[13] Surgical stabilization in pediatric patients may be warranted in the presence of a neurological deficit, when the atlanto-dens interval is increased to 8 mm, or when a marked ligamentous injury is suggested by anterior translation.[14] Osseous avulsion of the transverse ligament or occipital condyle fracture, with a small fragment of bone torn from C1 may be indicated by CT investigation[8] as seen in the present case (**Figures 1B, 2B**). Nonoperative management of this type of injury in a pediatric patient may result in an acceptable degree of stability.[8]

Conclusion

The current cases demonstrate that delayed spontaneous reduction of traumatic pediatric AARS is possible with conservative treatment. In the absence of compelling evidence to the contrary, a conservative approach may provide another option in the management of traumatic pediatric AARS. However, further studies with a larger number of cases may be necessary.

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