CASE REPORT

Not your typical torticollis: a case of atlantoaxial rotatory subluxation

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SUMMARY

A 9-year-old boy was pushed while carrying a heavy backpack. He had no fall or direct trauma to his neck. After a few hours from the incident, he presented with neck pain and torticollis. However, somewhat atypical for torticollis, he was not able to rotate his face to the neutral position. There were no neurological deficits. Concerned by his inability to rotate his neck, the clinician suggested CT of the upper cervical spine, which demonstrated rotary subluxation of the atlantoaxial junction. The patient was taken to the operating theatre. Reduction was performed under general anaesthesia. He did well and was back to school in the following days.

BACKGROUND

Atlantoaxial rotatory subluxation (AARS) is an uncommon but potentially life-threatening injury that presents as acute torticollis. We describe a case of AARS in a boy after a minor blunt trauma. It is an important differential that must be kept in mind while encountering paediatric cases of neck pain, especially in those patients who present with the typical 'cock-robin' posture.

CASE PRESENTATION

A previously healthy 9-year-old boy was brought to the emergency department with neck pain and rotation of his head to the right side with an inability to straighten it. He had acquired this injury a few hours previously, after he was pushed while carrying a heavy backpack. He had no fall or direct trauma to his neck. There was no loss of consciousness, headache or a recent history of upper respiratory tract infection. His medical history was unremarkable.

On examination, he was calm, cooperative and stable. His head was tilted to the right side while

Figure 2 Coronal view of CT of the cervical spine showing disrupted relation of C1 to C2.

being rotated towards the left. Slight anterior flexion of the head was also present. There was no tightness or tenderness over the sternocleidomastoid muscle. The ear, nose and throat examination was normal. There was no sign of any neurological deficit. Although the boy was very cooperative during the examination, there was painful restriction of even minimal movements of the head from the fixed position. Therefore, repositioning of the head was not attempted.

INVESTIGATIONS

With this clinical finding and given the history of mild trauma, the physician decided to opt for further imaging studies. The cervical CT scan showed type I AARS (figures 1–4).

Figure 1 Axial view of CT of the cervical spine showing atlantoaxial rotatory subluxation.

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Figure 3 Sagittal view of CT of the cervical spine showing forward stepping of C1 over C2.

Figure 5 Coronal view of CT of the cervical spine postreduction status which shows proper alignment and positioning of C1 in relation to C2.

DIFFERENTIAL DIAGNOSIS

Other causes of sudden onset acquired torticollis include atlanto-occipital subluxation, C1–C2 dislocation, cervical spine fracture, clavicle fracture, muscle injury/myositis, ligamentous injury, spinal epidural haematoma, respiratory infections, dystonia and drug-induced torticollis.

TREATMENT

The patient's family was offered a conservative approach and reduction under sedation. They opted to have reduction which was performed under sedation by our trauma team.

OUTCOME AND FOLLOW-UP

The patient did well after the reduction and there were no neurological complications. [Figure 5](#) shows the cervical spine CT scan after reduction. He was discharged with analgesia and soft cervical collar. He had a complete recovery and was back to school in the following days.

DISCUSSION

AARS is also known as atlantoaxial rotatory fixation. It is an uncommon but potentially life-threatening injury that presents

as acute torticollis.^{1 2} It can occur either spontaneously or as a consequence of trauma or congenital malformations of the spine.³ The mechanism of trauma is usually related to forced rotatory movements of the neck. AARS is a rotational disorder of the atlantoaxial joint that results in either limited rotation of the neck or, in rare instances, a complete fixation of the joint.³ It may or may not have a concurrent C1–C2 dislocation. The dire neurological complications of this injury are related to the proximity of the atlantoaxial joint to the medullocervical junction and adjacent vertebral arteries.

AARS has been classified into four types according to the radiological criteria specified by Fielding and Hawkins.⁴ This classification depends on the degree of displacement of the odontoid process of the axis relative to the anterior facet of the atlas. Type I is the most common subtype, which consists of rotatory subluxation without anterior displacement of the atlas (with the atlanto-odontal distance being ≤ 3 mm).

This injury is more often seen in children compared with adults.^{5 6} Adult AARS is often related to severe traumatic injuries and often results in neurological morbidity.⁷ However, incidence of neurological injury as a complication of AARS is fairly uncommon in paediatric AARS.⁸ In children, AARS is mainly related to

Figure 4 A three-dimensional reconstruction CT of the cervical spine confirming the C1 on C2 rotary subluxation.

inflammatory conditions of the head and neck, which contributes to increased laxity of the ligamentous structures in the atlantoaxial level, thus predisposing to a rotatory deformity.⁹ Certain other anatomical considerations in children such as the horizontal configuration of the C1–C2 articular facets, a rotational angle of greater than 45° and underdevelopment of neck musculature also make them more prone to AARS.⁷ Thus, they can sustain this injury even after trivial trauma, as illustrated in our case. Whether or not carrying a heavy backpack predisposed our patient to AARS is debatable. A heavy backpack would increase the inertia of the boy's trunk relative to the head. So a mild trauma can cause an inertial injury to the soft tissues of the vertebrae joints of the cervical neck, in which case, an even slight rotatory movement of the neck would make him prone to AARS.

The diagnosis of AARS requires high index of clinical suspicion along with thorough history and clinical examination. It must be differentiated from other more 'benign' causes of torticollis in children. CT scan of the cervical spine is recommended not only as a diagnostic tool but also for radiological staging of AARS.⁶ Dynamic CT with three-dimensional reconstruction is the best imaging modality for AARS.¹⁰

Prompt diagnosis of this condition is important as it facilitates timely management so as to prevent its serious long-term consequences.¹¹ Management of AARS depends on the duration and severity of the subluxation and is tailored according to the clinical scenario. Most cases can be managed by either immobilisation with a cervical collar or a combined strategy of traction

with immobilisation.^{2 8 12} A small retrospective study by Subach *et al* showed that in majority of cases of AARS that present early after subluxation, reduction can occur spontaneously with simple immobilisation. However, closed reduction with cervical traction, followed by rigid immobilisation, achieved reduction more rapidly and was successful in 94% of cases and curative in 63% of cases.⁸ Most children have excellent prognosis after reduction.

Most cases can be managed without operative intervention, producing excellent outcomes. Nevertheless, surgical intervention is usually reserved for cases that are complicated by neurological compromise, atlantoaxial instability or frequent recurrence.^{13–15}

Competing interests None.

Patient consent Obtained.

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Learning points

- ▶ Cases of acute painful torticollis are not always benign.
- ▶ In patients with a history of trauma various cervical spine injuries can present as torticollis.
- ▶ A diagnosis of atlantoaxial rotatory subluxation (AARS) should be considered in children who present with an inability to turn their head after minor trauma.
- ▶ In cases of torticollis following trauma, the assessment for involvement of the spinal cord should be given the first priority.
- ▶ In cases with AARS, a careful neurological examination should be conducted to determine the presence of any neurological compromise.

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