



Atlantoaxial rotatory subluxation in children: A review

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Abstract

Torticollis is a common complaint of childhood. Trauma should always be considered and carefully excluded. All children with torticollis should be examined with plain radiographs to rule out a fracture or bony abnormality. Atlantoaxial rotatory subluxation has been defined as a cause of temporary, self-resolving torticollis in children. However, on occasion it may be a potentially severe rotational deformity of the cervical spine. Early diagnosis of the lesion, proper evaluation and prompt treatment leads to a permanent resolution of the deformity, while misdiagnosis may lead to chronic deformity. In addition, fracture of the clavicle is one of the commonest injuries in childhood. However, torticollis associated with clavicular fracture is extremely rare. This co-existence in children should always be considered as a potential atlantoaxial rotatory subluxation. Two new cases as well as an extensive review of the literature are presented in this report.

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Review

Torticollis is seen at all ages, from newborns to adults, and refers to rotational deformity of the cervical spine with secondary tilting of the head. In children, it may be congenital or post-natally acquired. Acquired torticollis in children is a symptom that is usually due to a number of benign underlying lesions. However, severe and life threatening causes have also been encountered. The latter include musculoskeletal, ophthalmologic, infectious, neurologic, and neoplastic diseases that may exhibit an early presentation, with only torticollis [1-6].

Torticollis secondary to trauma is most commonly due to muscle spasm or injury. Physical examination is essential in the diagnostic investigation of patients with acquired torticollis resulting from trauma. Rare post-traumatic causes of acquired painful torticollis may include fracture or dislocation of the od-

ontoid, fracture of the first or second cervical vertebra, fracture of the first rib, atlantoaxial rotatory subluxation (or displacement) and a clavicular fracture [7-13].

Diagnosis of atlantoaxial rotatory subluxation is difficult and often delayed. The onset is spontaneous and usually occurs following minor trauma, or may follow an upper respiratory tract infection (Grisel syndrome) [14-18]. The child presents with torticollis and resists any attempt to move the head because of pain. The head is tilted to one side and rotated to the opposite side with the neck slightly flexed (a typical 'cock robin' position of the head). The associated muscle spasm is noted on the side of the 'long' sternocleidomastoid muscle because the muscle is attempting to correct the deformity, unlike congenital muscular torticollis, in which the spasmodic muscle causes the torticollis.



In acute cases attempts to move the head cause pain. Patients are able to increase the deformity but cannot correct it past the midline [19-26]. If the deformity becomes fixed (atlantoaxial rotatory fixation), the pain subsides but the torticollis persists along with diminished range of neck motion. In long-standing cases, plagiocephaly and facial asymmetry with flattening may develop on the side of the tilt [27-29]. Neurologic evaluation should carefully determine any neurologic compression or vertebral artery compromise [30].

Conventional radiography, including anteroposterior, lateral and odontoid views, should be the first-line imaging modality. Congenital anomalies and normal variants of the immature anatomy of the cervical spine should also be carefully defined in children suffering from torticollis after trauma [31]. The radiologist plays a key role in confirming the diagnosis [32].

Interpretation of radiographs is difficult. On the open mouth view there is loss of symmetry between the lateral masses of the atlas and the odontoid process. The lateral mass of the atlas that is rotated anteriorly appears wider and closer to the midline (medial offset), whereas the opposite lateral mass is narrower and away from the midline (lateral offset). The facet joints may be obscured because of apparent overlapping. The lateral view shows the wedge-shaped lateral mass of the atlas lying anteriorly, than the oval arch of the atlas normally lies, and the posterior arches fail to superimpose because of the head tilt. The normal relationship between the occiput and atlas is preserved. A lateral radiograph of the skull may show the relative position of cervical vertebrae 1 (C1) and cervical vertebrae 2 (C2) more clearly than a lateral radiograph of the cervical spine. Lateral flexion and extension views should be obtained to document any atlantoaxial instability [33]. In some children, the anterior physiological displacement of axis on the third cervical vertebra is so pronounced that it appears pathological (pseudosubluxation) [34, 35]. Swischuk has used the posterior cervical line, drawn from the anterior cortex of the posterior arch of atlas to the anterior cortex of the posterior arch of the third cervical vertebra, to differentiate it from pathological subluxation [36].

Acute post-traumatic torticollis is not necessarily the sign of a pathologic condition of the atlantoaxial joint. It is also not necessary to obtain computed tomography scans (static or dynamic) in this group of patients at the time of presentation. However, children presenting with resistant, unresolving torticollis may suffer from atlantoaxial rotatory subluxation.

Post-traumatic atlantoaxial rotatory subluxation is a rare, but potentially severe, cause of acquired torticollis in children. Atlantoaxial rotatory subluxation represents a wide spectrum of injuries. Atlantoaxial rotatory fixation may occur within the normal range of rotation of the atlantoaxial joint. In these cases, the joint is neither subluxed nor dislocated. The obstruction is probably due to capsular or synovial interposition. It may also be due to anterior shift of the atlas on the axis following fractures or ligamentous deficiency leading to atlantoaxial instability [37].

The increased incidence of atlantoaxial rotatory subluxation in children, compared to adults, may be related to certain anatomical differences. The dens-facet angle of the axis is steeper in children than in adults. Meniscus-like synovial folds are found in the first two cervical vertebrae facet joints of the spines in children, but not in those of adults [38].

The atlantoaxial rotatory displacement, as defined initially

by Fielding and Hawkins in 1977, is classified into four types: Type 1 is the most common form in children. It is a simple rotatory displacement without an anterior shift. Type 2 is potentially more dangerous. It is a rotatory displacement with an anterior shift of 5 mm or less. Type 3 is rotatory displacement with an anterior shift greater than 5 mm. Finally, type 4 is rotatory displacement with a posterior shift. Type 3 and 4 deformities are rare, but neurological involvement or even instant death may follow [39,40].

Diagnosis of the condition is largely clinical but may be aided by various imaging studies, including radiographs, static or dynamic Computed Tomography (CT) scanning, three-dimensional CT reconstructions, or magnetic resonance imaging. The radiological technique of choice for this condition is CT. Consideration should always be given to infection or other inflammatory disease as an underlying, precipitating cause [41-50].

Treatment includes observation if the complaints are mild and have been present for less than a week, short bed rest, a simple soft collar and analgesics. Most cases prove transitory and spontaneously resolving. Whenever the stiff neck and the slightly twisted head do not resolve in a few days, more aggressive treatment should be instituted. The most important factor for success of conservative treatment is the time from the onset of symptoms to recognition and initiation of treatment. In cases that reduction does not occur spontaneously or the rotatory subluxation is present for longer than 1 week, but less than 1 month, hospitalization and cervical traction are indicated. Head-halter traction is used, but halo traction may be required when torticollis persists for longer than 1 month [51-57].

Indications for operative treatment include neurologic involvement, anterior displacement, failure to achieve and maintain correction if the deformity exists for longer than 3 months, and recurrence of the deformity after an adequate trial of conservative management consisting of at least 6 weeks of immobilization [58-61]. If left untreated, persistent deformity due to the development of secondary changes in the bony anatomy of the atlantoaxial joint may be evident [62].

Pediatric emergency physicians should have a high clinical suspicion for atlantoaxial rotator subluxation, particularly when a child presents with neck pain and an abnormal head posture without the ability to return to a neutral position [63].

The diagnosis of atlantoaxial subluxation should always be considered in children presenting with a clavicular fracture associated with acute torticollis. It has been postulated that the rotary displacement is a direct result of the traumatic injury that produces the fracture. The head is most often laterally bent toward and rotated away from the fractured clavicle. Treatment of the clavicle fracture is straightforward, but failure to recognize and treat the atlantoaxial rotatory subluxation promptly may lead to a fixed deformity [64-70].

Cases with a diagnosed atlantoaxial rotatory subluxation associated with a fractured clavicle, although very rare, have sufficiently been discussed in the world literature. However, reports on missed cases and their final outcome are not evident.

We were able to identify, from the hospital database, two cases with atlantoaxial rotatory subluxation.

The former was a 10-year-old boy that presented with acute torticollis and neck stiffness after falling on backwards a day ago. The frontal and open mouth odontoid views demonstrated

atlantoaxial rotatory subluxation (Figure 1A). On the lateral radiograph loss of the normal lordosis was only diagnosed (Figure 1B). Diagnosis was clearly secured with the CT scan findings. Coronal (Figure 1C), sagittal (Figure 1D) and axial (Figure 1E) views showed that the odontoid was lying eccentrically between the lateral masses of the atlas. A type I simple atlantoaxial rotatory displacement was diagnosed. The neck symptoms were considerably relieved immediately after gentle manipulative axial traction.

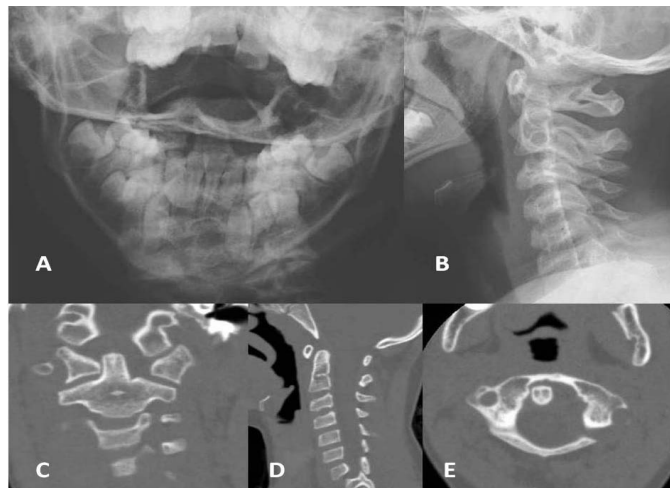


Figure 1: Initial anteroposterior radiograph of the cervical spine indicated loss of symmetry between lateral masses of the atlas and odontoid process (A). Lateral cervical spine radiograph showed loss of the normal lordosis (B). Coronal CT images demonstrated narrowing of the right lateral atlantodental interval and a wider left lateral atlantodental interval (C). Sagittal CT views of the cervical spine indicated no abnormal findings (D). Axial CT views confirmed that the odontoid was lying eccentrically between the lateral masses of the atlas (E).

The latter was an 8-year old boy that presented with torticollis associated with an injury of the left shoulder. Plain anteroposterior (Figure 2A) radiograph showed a midshaft fracture of the left clavicle, while the lateral view was indicative of a reversed cervical lordosis (Figure 2B). The potential severity of the cervical spine injury was overlooked. No CT scanning evaluation was recommended. Bed rest and analgesics was the offered treatment. The patient reported an uneventful recovery of the torticollis within a few days.

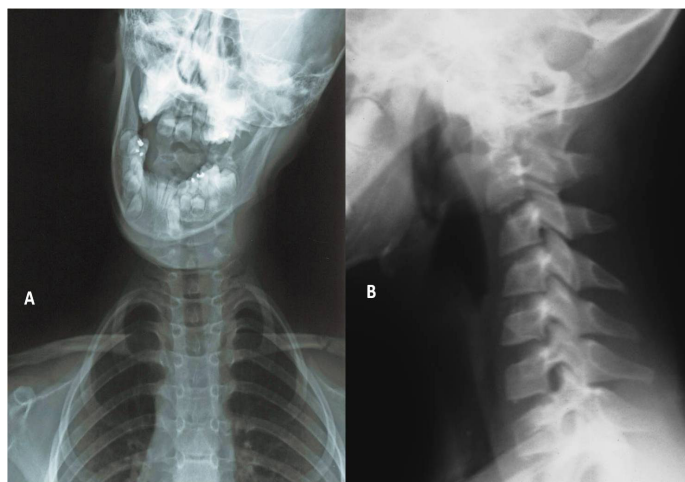


Figure 2: Typical torticollis position of atlantoaxial rotatory displacement. The head is tilted to the side of the fractured clavicle and rotated to the opposite side, with slight flexion. Associated muscle spasm, unlike muscular torticollis, is predominantly on the side of the 'long' sternocleidomastoid, because the muscle is attempting to correct the deformity. Initial anteroposterior radiograph of the cervical spine, demonstrating left lateral cervical tilt due to the midshaft clavicular fracture (A). On the lateral projection no abnormal alignment of the atlas and the axis was evident but there was a reversed cervical lordosis. The anterior displacement of the axis on the third cervical vertebra was not considered pathological (B).

Conflict of interest statement

The author certifies that he has no commercial associations (such as consultancies, stock ownership, equity interest, patent/licensing arrangements, etc) that might pose a conflict of interest in connection with the submitted article.

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