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Delayed closed reduction of rotatory atlantoaxial dislocation in an adult

Received: 22 May 2000
Revised: 20 October 2000
Accepted: 31 October 2000
Published online: 29 June 2001
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Abstract We report a case of rotatory atlantoaxial dislocation due to a rugby injury in an adult. The patient presented with torticollis 4 weeks after the injury. The neurological evaluation was normal. Reduction proved difficult to obtain and required 10 days of skull traction followed by gentle manipulation. After reduction, dynamic cervical radiographs showed no instability and magnetic resonance imaging (MRI) confirmed that the transverse ligament was intact. After 6 weeks of immobilization in a Minerva jacket, a dynamic rotatory computed tomography (CT) scan confirmed that the atlantoaxial joint was stable. Such

cases of atlantoaxial joint dislocation in adults treated by traction after a considerable delay are rare. In our patient, demonstration by MRI that the transverse ligament was intact led to the decision to use conservative therapy, which proved successful.

Keywords Atlantoaxial joint · Dislocation · Sports-related injury · Traction · Magnetic resonance imaging

Introduction

Rotatory dislocation is an abnormality seen in the upper cervical spine, where most of cervical rotation occurs [11]. Rotatory atlantoaxial dislocation was first reported in children [12] and is uncommon in adults, most cases being seen after severe trauma [3]. A classification scheme was developed by Fielding and Hawkins [12]. Rotatory atlantoaxial dislocation can result in tetraplegia due to proximal spinal cord compression [17] or in upper limb deficits due to nerve root compression [3]. Factors that influence therapeutic choices include time since injury, ease of reduction, presence of neurological compromise, and stability of the lesion.

Case report

A 41-year-old man reported that, while scrimmaging during a rugby match 1 month earlier, he had received an injury that twisted

his neck and produced a cracking sound. He immediately experienced a sensation of heaviness and weakness in both upper limbs, with no paresthesia. Spinal muscle guarding caused torticollis. At examination, there was no neurological deficit, and immobilization by a cervical collar was deemed sufficient.

He was referred to our department 1 month after the injury because of persistent torticollis. His neck was fixed in lateral flexion and rotation toward the left. There was no sign of spinal cord or nerve root compromise or of pyramidal tract irritation. Anteroposterior open-mouth radiography (Fig. 1) and lateral radiography of the cervical spine (Fig. 2) demonstrated rotatory atlantoaxial dislocation. A computed tomography (CT) scan with reconstruction confirmed the presence of rotatory dislocation (Fig. 3, Fig. 4). The rotation axis was the dens, and the atlas–dens interval was normal. These findings established the diagnosis of rotatory atlantoaxial dislocation corresponding to type 1 in the classification scheme developed by Fielding and Hawkins [12].

Reduction of this 4-week-old dislocation proved difficult. Skull traction was started immediately at 3 kg and was gradually increased to 6 kg. Cranial nerve function was closely monitored. Although muscle relaxants and analgesics were given, reduction proved difficult, and the possible need for open reduction via the transoral route was discussed with the patient. However, reduction was obtained after 10 days by manipulation of the neck. The re-

Fig. 1 Anteroposterior open-mouth radiograph showing the cervical spine under the dislocation as if viewed from an oblique view

Fig. 2 Lateral radiographs of the cervical spine. C1 is viewed laterally and the rest of the cervical spine appears in a three-quarters oblique view

duction maneuver consisted in increasing the distraction by pulling gently on the neck at the bedside, while the patient was conscious, then in a movement aimed at correcting the lateral flexion and rotation. This movement corrected the torticollis, at the same time producing neck pain. Anteroposterior open-mouth radiographs confirmed that the dislocation had been reduced. To determine whether C2–C1 fusion was required, dynamic radiographs of the cervical spine were obtained (Fig. 5). There was no evidence of instability, and the atlas–dens interval was normal (C1–C2, <3 mm). A magnetic resonance imaging (MRI) study was performed to look for post-traumatic spinal cord contusion and, above all, to evaluate the transverse ligament. The findings (Fig. 6) established that the transverse ligament was intact. We therefore decided to employ

Fig. 3 Computed tomography findings of rotatory dislocation of both atlantoaxial joints. The axis of rotation is the dens

Fig. 4 Three-dimensional reconstruction showing rotatory dislocation of both atlantoaxial joints

conservative treatment using a Minerva jacket with an occipital and a chin rest.

The Minerva jacket was removed after 6 weeks. Neurological findings were normal. Rotation of the neck was 10° toward the right and 30° toward the left. The range of motion of the neck in the other planes was normal. A dynamic CT study obtained in forced rotation confirmed that the atlantoaxial joint was stable: in

Fig. 5 Dynamic radiographs of the cervical spine: atlas–dens interval less than 3 mm, no forward slippage of C2 on C3

Fig. 6 Magnetic resonance imaging (MRI) scan showing that the transverse ligament is intact

maximum rotation, the atlantoaxial angles were 9° and 10° on the right and left, respectively (Fig. 7). Rehabilitation therapy aimed at recovering mobility was started. Twelve months later, the range of motion of the neck was normal in all planes, and a dynamic lateral radiograph of the cervical spine confirmed that the atlantoaxial joint was stable.

Discussion

Rotatory atlantoaxial subluxation is common in children after minor injuries [9], as a manifestation of Grisel's syndrome [15], or after severe trauma (e.g., due to a motor vehicle accident) [22]. It is rare in adults. Torticollis is the presenting syndrome, and the dislocation can be missed [2, 15, 25]. CT increases the likelihood of making the diagnosis [2, 19, 30] and, when images in rotation are obtained, is useful for evaluating reducibility of the lesion [2, 14, 26]. Fielding and Hawkins [12] distinguish four stages based on the extent of the shift of C1 on C2: *type 1*

Fig. 7 Follow-up computed tomography study in rotation, with measurement of the C1–C2 angles. **A** Right rotation. **B** Left rotation

is characterized by bilateral rotatory dislocation around the dens with a normal atlas–dens interval; *type 2* by unilateral rotatory dislocation around one of the lateral masses with a 3- to 5-mm atlas–dens interval, indicating the presence of transverse ligament disruption; *type 3* by a greater than 5-mm atlas–dens interval, indicating disruption of both the transverse ligament and the alar ligaments; and *type 4* by posterior displacement of C1 due to failure of the dens.

As in all dislocations, the treatment consists in immediate reduction. If the trauma was minor, a collar, cervical traction [11, 15], or manipulation [10] may allow reduction of the dislocation to be obtained. However, the success rate of conservative treatment drops if the diagnostic delay exceeds 1 month [26]. A severe trauma responsible for transverse ligament disruption can cause a combination of sagittal and rotatory dislocation. According to Fielding and Hawkins [11], an atlas–dens interval greater than 5 mm in children or 3 mm in adults indicates transverse ligament disruption, a lesion associated with a risk of spinal cord compression [8, 13]. The choice between conservative treatment and C2–C1 fusion is directly de-

pendent on whether or not the transverse ligament is torn or avulsed [8, 16]. C2–C1 instability due to severe trauma requires fusion and, if neurological compromise is present, spinal cord decompression [13, 22].

Few cases of rotatory atlantoaxial dislocation have been reported in adults [3, 4, 20, 24, 27, 28], perhaps because the cause may often be severe trauma responsible for lethal injuries [6, 24]. Jones reported a case of rotatory dislocation of both atlantoaxial joints due to an injury sustained in a rugby scrimmage [20] and responsible for hemiparesis. The two patients with atlantoaxial dislocation reported by De Beer et al. [6] were also injured during a rugby match. In our patient, an injury sustained during a rugby scrimmage caused transient incomplete diplegia of the upper limbs. The mechanism of spinal cord compression in rotatory atlantoaxial dislocation was explained clearly by Mazzara and Fielding [23], who showed that bilateral dislocation occurs at 63° of rotation and reduces the spinal canal diameter by 1 cm, thus potentially causing spinal cord compression followed by myelomalacia. When the diagnosis is delayed, reduction of the dislocated atlantoaxial joint can be extremely difficult, requiring several weeks of skull traction without overdistractive in unstable lesions [6]. If this fails, open reduction via the transoral [21] or retrosternomastoid [5] route must be performed. Several studies [4, 14] have convincingly demonstrated the usefulness of CT for visualizing the dislocation, determining whether it is unilateral or bilateral, and looking for fractures. However, the choice between conservative therapy and fusion depends on whether or not the lesion is stable. The transverse ligament is intact in patients with bilateral rotatory atlantoaxial dislocation, but is ruptured in unilateral dislocation [1, 28]. The transverse ligament is a primary stabilizing component of the atlantoaxial joint. On lateral radiographs of the cervical spine in flexion and extension, an atlas–dens interval (i.e., the distance between the anterior arch of C1

and the dens) of less than 3 mm indicates that the transverse ligament is intact and the atlantoaxial joint stable. MRI directly visualizes the transverse ligament [7] as a line of low signal on T₂-weighted images uniting the two lateral masses of C2 anterior to the high signal generated by the cerebrospinal fluid. MRI allows detection of a tear in the midportion of the transverse ligament in the absence of avulsion of a bony fragment. An intact transverse ligament is an argument in favor of conservative therapy [2]. High success rates have been reported with conservative therapy in patients with promptly diagnosed, stable, rotatory atlantoaxial dislocation [18, 20, 24] not associated with transverse ligament disruption. In patients with instability due to transverse ligament disruption, C2–C1 fusion by screws is the option associated with the smallest degree of motion range limitation [3, 29].

Conclusions

Rotatory atlantoaxial dislocation is relatively rare in adults but can be caused by neck injuries during sporting activities such as rugby. Torticollis due to muscle guarding is the presenting symptom, and the diagnosis is easily missed if the neurological evaluation is normal. An anteroposterior open-mouth plain radiograph suggests the diagnosis, which can be confirmed by CT. Reduction is difficult if the diagnosis is delayed by more than 1 month. Skull traction is indicated, if necessary in association with direct manipulation of the neck. MRI shows the transverse ligament as a low signal on T₂-weighted images. Stability is dependent on whether the transverse ligament is intact and can be evaluated by measuring the atlas–dens interval on dynamic cervical radiographs and the atlas–axis angle on CT scans in maximum rotation. In stable lesions, conservative therapy with a Minerva jacket produces good results.

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