CASE REPORT

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Post-traumatic Atlantoaxial Rotatory Dislocation in an Adult Treated by Open Reduction and C1-C2 Transpedicular Screw Fixation

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Atlantoaxial rotatory dislocation (AARD) is an uncommon disorder of childhood in which clinical diagnosis is generally difficult and often made late. It is very rare in adults because of the unique biomechanical features of the atlantoaxial articulation. We report a case of post-traumatic AARD in an adult. Reduction was difficult to obtain by skull traction and gentle manipulation. Therefore, the patient was treated surgically by an open reduction, transpedicular screw fixation, and posterior C1-2 wiring with graft. The normal atlantoaxial relation was restored with disappearance of torticollis. Postoperatively, the patient remains neurologically intact and has radiographic documentation of fusion. Atlantoaxial transpedicular screw fixation can be one of the treatment options for the AARD.

KEY WORDS : Atlantoaxial dislocation · Rotatory · Transpedicular screw.

Introduction

Atlantoaxial rotatory dislocation (AARD) is an uncommon disorder of childhood in which resolution usually occurs spontaneously or after traction therapy[8]. In a minority of children, irreducible or chronic fixation may develop and the natural history then usually involves restriction of head on neck movement, abnormal head position, and progressive facial asymmetry[15]. Unlike to AARD in children, post-traumatic AARD is very rare in adults due to the unique biomechanical features of the C1-C2 articulation and the probable lethality of injury to the adjacent medulla or vertebral arteries before presentation at the trauma center[3,12].

We present a rare case of type II post-traumatic AARD in an adult successfully treated with open reduction and transpedicular screw fixation with posterior wiring.

Case Report

A 34-year-old male patient was referred to our department after a fall from stairs a day before admission. His chief complaint was pain in left upper cervical area. Neurological examination is normal but his head was fixed and rotated to right side and tilted toward left side with decreased neck motion. Radiographs taken in flexion and extension and open mouth view showed 4mm of atlantodental interval (ADI) and left

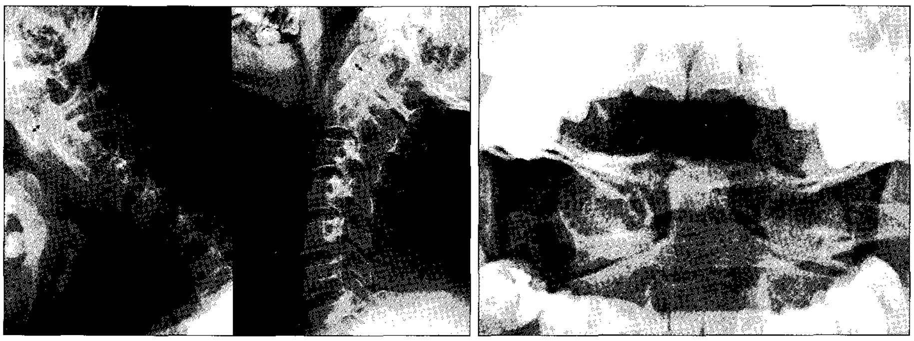

Fig. 1. Preoperative flexion and extension view demonstrating 4 mm of atlantodental interval (arrows) and open mouth view revealing left C1 lateral mass more closer to the midline than right one.

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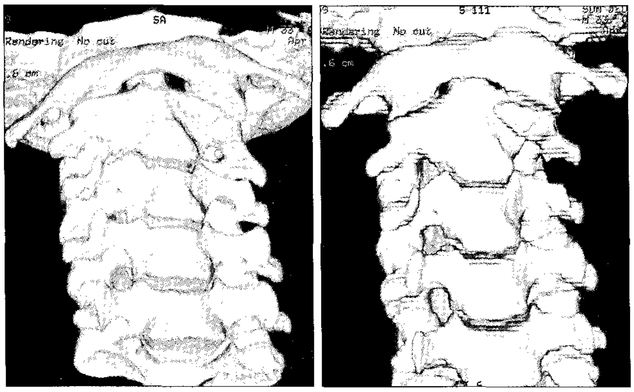

Fig. 2. Cervical spine computed tomography with three-dimensional reformations revealing anterior subluxation of left lateral mass of C1 with respect to C2, right lateral mass of C1 acting as the pivot point for rotation, and additionally, fracture of left superior articular facet of C2.

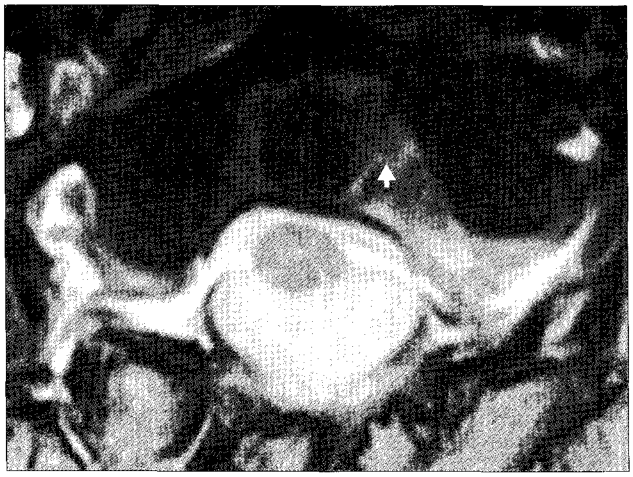

Fig. 3. The axial T2-weighted image of cervical spine magnetic resonance image revealing mild deficiency of left-side transverse ligament (arrow).

C1 lateral mass more closer to the midline than right one (Fig. 1). Cervical spine computed tomography (CT) with three-dimensional reformations revealed anterior subluxation of left lateral mass of C1 with respect to C2, right lateral mass of C1 acting as the pivot point for rotation, and additionally, fracture of left superior articular facet of C2 (Fig. 2). Evidence of traumatic injury of both vertebral artery was absent in neck CT angiography. The axial T2-weighted image of cervical spine magnetic resonance image (MRI) revealed partial injury of left-side transverse ligament (Fig. 3). Closed manual reduction in supine position was tried by compression of anteriorly, dislocated, left C1 arch with hands under Gardner traction, but failed. Subsequently, open reduction in prone position was tried under general anesthesia. After exposure of C1-2 posterior arch, Songer cable was passed under the left arch of C1 and then the wire was pulled upwardly with Gardner traction. Successful reduction was done with audible "pop" sound. After detection of medial margins of C1, C2 pedicles, 28 mm-long, 4 mm-diametered screws were inserted via pedicular

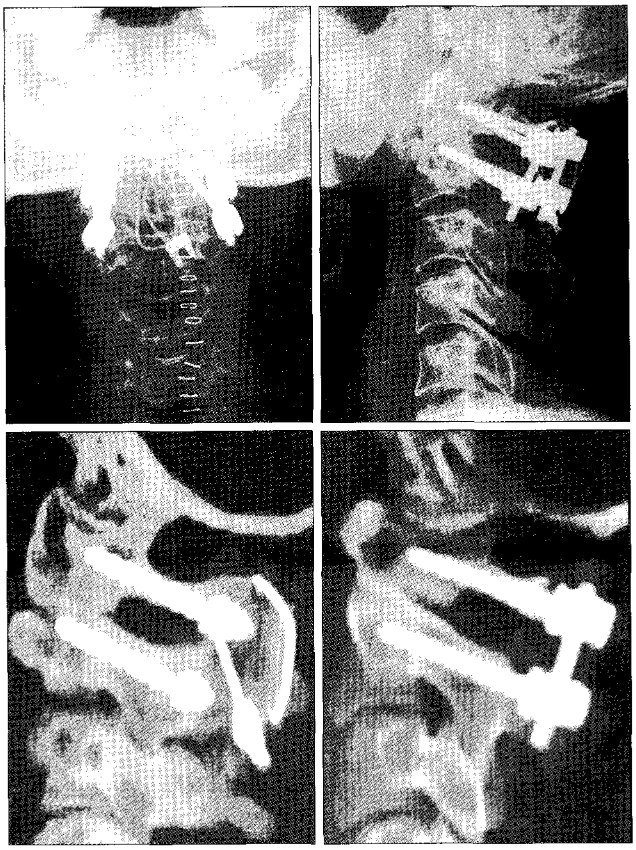

Fig. 4. Postoperative simple radiographs and sagittal reconstruction images of computed tomography showing a good position of the C1-2 complex and a correct insertion of screws through C1-2 joints.

entry point with the trajectory 10 degrees medially in C1 and 20 degrees medially and superiorly toward superior-medial quadrant of C2 lateral mass in C2. C1-2 screws were fixed by 30 mm rods with compressive force. Then, posterior C1-2 wiring was done with autologous iliac bone graft by Sonntag

Fig. 5. Lateral radiograph at 1 year follow-up demonstrating bone bridging between C1-2 arches and a atlantoaxial stable fusion.

method. Postoperative course was uneventful without any complications, and posterior cervical pain and torticollis were relieved immediately. He was able to ambulate well with soft color brace from the next day after the operation and postoperative radiographs and CT showed a good position of the C1-2 complex and a correct insertion of screws through C1-2 joints (Fig. 4). Lateral roentgenograms at one year follow-up demonstrated bone bridging between C1-2 arches and a atlantoaxial stable fusion (Fig. 5).

Discussion

AARD represents a rare pathological condition of the upper cervical spine that is frequently misdiagnosed, leading to a delay in therapy[8]. The cause of AARDs is usually either an infection or a traumatic event, but the condition may also arise spontaneously or in association with other conditions. Isolated case reports have documented rotatory deformities caused by ankylosing spondylitis[13], metastatic tumor[25], generalized ligamentous laxity[15,27], eosinophilic granuloma[15] following a suboccipital craniotomy and C1-3 laminectomy[23], and various other non-destabilizing procedures about the head and neck[26].

The pathophysiology of AARDs is not well defined, but there is a clear predilection for children and young adults regardless of the causes[8]. The higher incidence of AARD in children is the consequence of the specific anatomical features[16]. The joint surface of the lateral mass is shallower and more horizontally oriented. In addition, the relative elasticity of the ligaments, the not yet fully developed neck muscles, and the relatively large head might be predisposing factors for AARD. Isolated rotatory dislocation is very rare in adults[9,10,18]. Few cases of AARD have been reported in adults, perhaps because the cause may often be severe trauma responsible for lethal injuries[1-3,10,12,17,19,20,22].

In the typical AARD, the patient's head is rotated away from the anteriorly displaced C1-2 joint and tilted toward the involved side. On palpation, the C2 spinous process may be prominent and deviated to the side to which the chin is pointed, as a result of the lateral tilt of the head or possibly from counterrotation of C2 in an attempt to realign the head[8]. Neurological involvement is fortunately uncommon and occipital neuralgia may occur because the greater occipital nerve runs in close proximity to the C1-2 facet capsule[5,8].

The atlantoaxial joint is stabilized by the transverse ligament and the alar ligaments. The transverse ligament prevents excessive anterior shift of the atlas on the axis, whereas the alar ligaments prevent excessive rotation; the right alar ligament limits left rotation and vice versa[5]. The transverse ligament may be incompetent, leading to a widened ADI on lateral radiographs or CT scan. On open mouth view, the lateral mass of the atlas that is rotated anteriorly appears wider and closer to the midline than its counterpart on the opposite side, and the C1-2 joint spaces appear asymmetrical, often leading to a narrow joint space on one side and a widened space on the opposite side[8]. Computed tomographic (CT) scanning is an excellent method of demonstrating abnormal C1-2 relationships[18,21,28], visualizing the dislocation, determining whether it is unilateral or bilateral, and looking for fractures[2,3], and may also be used to demonstrate compensatory occiput-C1 rotation or the unusual occurrence of simultaneous subluxation of both occiput-C1 and C1-2 joints[18]. Three-dimensional reformations give a global view of the cervical deformity[2]. Direct visualization of the transverse ligament with MRI can also be possible. The normal ligament has a homogeneous low signal intensity on gradient-echo images, whereas loss of continuity of the ligament with regions of high signal intensity characterize a tear[7].

Fielding and Hawkins[8] described four types of AARD based on the extent of the shift of C1 on C2 and the integrity of transverse ligament, which have been widely accepted. Type I is the most common pattern and the transverse ligament is intact. The fixed C1-2 rotation is symmetrical and within the normal range of rotation for the C1-2 joints. Type II deformity is identified by mild deficiency of the transverse ligament with an ADI of 3 to 5 mm. The intact joint acts as the pivot point for anterior unilateral displacement of the opposite side. The amount of fixed rotation exceeds the normal excursion of the C1-2 complex. Type III deformity is identified by a greater than 5mm ADI, indicating disruption of both transverse ligament and alar ligament. Both lateral masses are displaced anteriorly, and one side is rotated further forward than the other. Type IV lesion is described as a posterior shift of one or both lateral masses of the atlas. The importance of the classification scheme by Fielding and Hawkins lies in the increasing risk of spinal instability with potential neurological compromise and the higher likelihood of recurrent deformity with the more severe types. In our case, the lesion was consistent with type II AARD by detailed radiological evaluation.

The goals of treatment are to restore normal pain-free range of motion, to prevent or reverse neurological compromise, and to restore spinal stability. If the trauma was minor, a collar brace, cervical traction, or manipulation may allow reduction of the dislocation to be obtained[21]. A surgical approach is advised for cases of AARD with spinal instability, neural involvement, or failure to achieve or maintain reduction by conservative measures[8,14,17,21]. Levine and Edwards[14] recommend performing the manipulation with the patient awake using topical anesthetic in the posterior pharynx. Gentle traction is applied through Gardner-Wells tongs, and the skull and C1 are derotated. They note that the reduction is often accompanied by an audible "pop" and can be confirmed by palpation of the ring of C1 through the mouth. If a closed reduction is unsuccessful, open reduction must be performed[6,24]. A wire passed under the arch of C1 can be used to manually derotate the atlas. After the deformity is reduced, the wire may be used to maintain the alignment by incorporating it into a C1-2 fusion[14]. In our case, manipulation was unsuccessful and derotation was achieved by open reduction ultimately. The choice between conservative treatment and C1-2 fusion is directly dependent on whether or not the transverse ligament is torn or avulsed. An intact transverse ligament is an argument in favor of conservative therapy[24]. When C1-2 fusion is indicated, a posterior fusion is considered as the procedure of choice. Among the various types of posterior fusion techniques, C1-2 transpedicular screw fixation has several advantages over other posterior fusion techniques including transarticular screw fixation[4]: 1) exposure of C1-2 joint or opening of C2 pars interarticularis is not necessary, so this procedure shortens the operative time, prevents troublesome venous bleeding, and decreases blood loss; 2) there is no postoperative discomfort secondary to pain and numbness involving the posterior scalp because there is no need to sacrifice the bilateral C2 dorsal root ganglia in this procedure; 3) this procedure can lessen the risk of injury to vertebral artery and spinal cord; 4) this procedure can provide immediate rigid internal fixation, permitting early mobilization and minimal external support. According to the recent reports on biomechanical comparison of fusion techniques, the posterior C1-2 fusion by transpedicular screws demonstrated the greatest biomechanical stiffness[24]; 5) this procedure can be one of the treatment options for atlantoaxial instability with bilateral high-riding vertebral artery[11]. On the basis of these factors, we performed C1-2 transpedicular screw fixation and posterior wiring to mild deficiency of the transverse ligament (Type II AARD) accompanied by facet fracture of C2, without any intraoperative complications.

Conclusion

In general, surgical fusion techniques is mandatory in patients with type II and more severe types of AARD due to C1-2 instability from the transverse ligament injury.

We report a rare case of type II post-traumatic AARD in an adult treated by open reduction and C1-2 transpedicular screw fixation with posterior wiring, without any surgical complications.

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