## CASE-BASED UPDATE

# Severe spinal cord injury in craniocervical dislocation. Case-based update

Juan F. Martínez-Lage • Fernando Alarcón • Raul Alfaro • Amparo Gilabert • Susana B. Reyes • María-José Almagro • Antonio López López-Guerrero

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## Abstract

*Background* Craniocervical distraction injuries, including atlanto-axial dislocation (AAD) and atlanto-ocipital dislocation (AOD), are often associated with severe spinal cord involvement with high morbidity and mortality rates. Many patients with these injuries die at the accident scene, but advances in emergency resuscitation and transport permit that many patients arrive alive to hospitals.

*Discussion* Children with craniocervical distraction injuries usually present with a severe cranioencephalic traumatism that is the most relevant lesion at admission. After resuscitation and hemodynamic stabilization, the spinal cord damage appears as the main lesion. Apnea and quadriparesis, or quadriplegia, are usually present at the onset. Early diagnosis and management perhaps decrease life-threatening manifestations of the spinal lesion. But even so, the primary

J. F. Martínez-Lage (⊠) · M.-J. Almagro · A. L. López-Guerrero Unit of Pediatric Neurosurgery, Regional Service of Neurosurgery, Virgen de la Arrixaca University Children's Hospital, 30120El Palmar, Murcia, Spain e-mail: juanf.martinezlage@cablemurcia.com

F. Alarcón · R. Alfaro
Unit of Spine Neurosurgery, Regional Service of Neurosurgery, Virgen de la Arrixaca University Hospital, 30120El Palmar, Murcia, Spain

A. Gilabert
Section of Pediatric Radiology, Virgen de la Arrixaca University Children's Hospital,
30120El Palmar, Murcia, Spain

S. B. Reyes Pediatric Intensive Care Unit, Virgen de la Arrixaca University Children's Hospital, 30120El Palmar, Murcia, Spain spinal cord insult is often irreversible and precludes obtaining a satisfactory functional outcome.

*Patients and methods* We report the findings of four children with craniocervical distraction injuries (AOD and AAD) who presented with severe spinal cord damage. All patients were admitted with respiratory distress or apnea together with significant brain injuries. The medical records pertaining to these patients are summarized in regard to clinical features, management, and outcome.

*Conclusions* In spite of timely and aggressive management, craniocervical injuries with spinal cord involvement continue to have a dismal prognosis. Outcome is closely related to the severity of the initial brain and spinal cord damage and is nearly always fatal in cases of complete spinal cord transection. Priority should be given to life-threatening complications. Ethic issues on indications for surgery deserve a detailed discussion with the children's parents.

**Keywords** Atlanto-axial luxation · Atlanto-occipital distraction injuries · Cervical spine fusion · Cervical spine injuries · Pediatric spinal cord injuries · Post-traumatic apnea · Quadriplegia

## Introduction

Osteoligamentous injuries at the craniocervical junction, although infrequent, may be life threatening. Distractive forces, as violent hyperflexion or hyperextension, with or without rotational movements of the neck, are apt of causing the most severe lesions. The peculiarities of the craniocervical junction of children may worsen the effects of injuries to this area. The consequences of these traumatisms in otherwise healthy patients may be catastrophic from the physical, economic, and moral viewpoint. In this work, we aimed at reporting clinical and neuroradiological features of children with atlanto-occipital dissociation (AOD) or atlanto-axial dislocation (AAD), remarking the diagnostic difficulties encountered at admission in the patients that survive the accident. Many patients arrive to hospital in coma, with hemodynamic instability and breathing difficulty that divert the attention to the most apparent lesion, usually a head injury.

## Pediatric spinal injuries

Osenbach and Menezes [1] reviewed 179 children with spinal traumatisms reporting an incidence of 9 % of all spinal injuries seen at their institution, which is within the range of other reported series that vary from 1 to 10 % [2–4]. The authors remarked the differences in the lesions encountered in children in relation with those of adolescents and adults. Anatomic-physiologic peculiarities of young children comprise (a) a proportional larger head size, (b) displacement of the fulcrum of motion rostrally (C2-C3), (c) horizontal disposition of the articular facets, (d) wedgeshaped vertebral bodies, and (e) immaturity and laxity of ligaments and muscles [1]. Most spinal injuries in children occur at the cervical region [1-5]. Of a total of 112 (62 %) neck injuries, upper cervical and craniovertebral injuries were more frequent in young children (36 %) than in older children and adolescents (26 %). Thoracic and thoracolumbar lesions accounted for 24 % and lumbar injuries occurred in 14 % of cases [1].

Vehicular accidents and falls were the most repeated cause of injury accounting for 56 and 17 %, respectively [1]. Complete spinal cord lesions were present in 23 % and incomplete cord lesions in 29 % of cases, while 48 % of patients retained an intact neurological function [1]. The distribution of lesions according to neuroimaging findings was fracture in 42 %, fracture/subluxation in 28 %, subluxation in 11 %, and spinal cord injury without radiographic abnormality (SCIWORA) in 19 % [1]. Hamilton et al. reviewed 174 hospital admissions of pediatric spinal injury reporting similar results [2]. In a companion paper, these authors analyzed 61 deaths associated with pediatric spinal injuries giving a mortality rate of 28 % [3]. Thirty-one children were submitted to a complete autopsy including 24 patients who died at the accident scene and seven deaths that occurred after hospital admission [3]. Death was directly due to the spinal cord injury in only eight patients, six instances with AOD and two with AAD [3]. In the other 22 children, the reported causes of death were massive hemorrhage (n=10), severe traumatic brain injury (n=7), and serious multiple traumatisms (n=5).

Martin et al. recently reported on the patterns in pediatric spinal trauma [4]. The authors reviewed a database comprising 19,538 instances of trauma that included 527 (2.7 %) of fracture/luxation without spinal cord injury and 109 (0.56 %)

with cord involvement. Cord injury and SCIWORA occurred more often in children aged  $\leq 8$  [4]. The risk of cord involvement increased with reduced GCS, head injury, and chest injury [4]. Ruge et al. reported data pertaining to 71 children, nine of them  $\leq 3$ years of age, and showed that the most frequent level of injury was C2 (15 cases, 27 %) [5].

## **Pediatric cervical lesions**

Approximately, 60 % of all pediatric spine injuries occur at the cervical region. The majority of cervical lesions at this age occur at the occipitocervical region and between C1 and C3 [1-4, 6-10]. The mechanisms involved in the production of injuries of the craniocervical and upper cervical spine are usually complex and they often occur in combination [6-10]. Flexo-extension and distraction-rotation displacements explain the majority of lesions in this area. Several series have dealt with the most habitual types of pediatric neck injuries [6-10]. Special head and neck injuries have been reported in abusive head trauma (n=52) and in car occupants without a head impact, both with a high mortality rate [11, 12]. Brennan et al. have documented neck injuries in 52 children that are victims of abusive head trauma that included 41 deaths [11]. Of these, 29 (71 %) had spinal cord injuries including 14 cases with associated brainstem injuries. No child had a vertebral fracture [11]. Huelke et al. performed a literature review of cervical spine injuries without a head impact in restrained occupants stressing the low frequency (0.4 %) of fractures and fracture dislocations found in this mechanism [12].

#### Atlanto-occipital and atlanto-axial dislocation

The skull base is connected with the upper cervical spine by the tectorial membrane, anterior and posterior atlantoocipital membranes, alar and apical ligaments, the cruciate ligament, and the joints between the occipital condyles and the lateral masses of the atlas [13–18]. All the aforementioned structures are often involved in AOD and AAD [13].

AOD has been rarely documented [13–18]. Some publications are case report studies, and many refer mainly to survivors of accidents [15–18]. Horn et al. documented 33 instances of AOD, including five deaths of children with severe head injuries who were not treated and 28 treated patients [13]. Five of the 28 treated children subsequently died [13]. This series also supported that most instances of AOD are unstable and need surgical fixation [13]. In the series of Cooper et al., of 69 patients with craniocervical dissociation, 47 were diagnosed post mortem, 22 were diagnosed in hospital, and only 7 survived to discharge [14]. AAD may be anterior, posterior or rotatory. Horizontal AAD can result from trauma, congenital conditions such as Down and Morquio syndromes, degenerative, and from inflammatory conditions such as rheumatoid arthritis [19–22]. Rotatory AAD has been recognized as an especial entity and has been discussed separately [19, 23]. An inflammatory type of C1–C2 subluxation is known as Grisel's syndrome [24]. Vertical AAD is more rarely reported [19].

Clinically, the patients with craniocervical distraction injuries may present either with minor manifestations as neck pain, cervical rigidity, mild myelopathy, or C2 hypesthesia, or they may present with an associated severe head injury that is often aggravated by hypoxic encephalopathy. In addition, these injuries may cause the immediate patients' death or produce a complete spinal cord transection with quadriplegia and apnea. Formerly, patients with significant dislocations of the occipitocervical and atlanto-axial segments died at the accident scene but present availability for resuscitation measures and for emergency transportation allow their survival and hospital admission.

Radiographic anatomy and diagnostic criteria in AOD and AAD have been dealt with extensively elsewhere [25–27]. Table 1 shows some neuroimaging findings in craniocervical injuries. Deliganis et al. [25] described the features that characterize patients at high risk for these injuries that comprise: (a) high-speed motor vehicle accident; (b) a crash resulting in death at the scene of the accident; (c) a fall from a height higher than 2.5 m, significant head injury, or evidence of intracranial hemorrhage in CT; (d) neurologic signs or symptoms referred to the cervical spine; and (e) pelvic or multiple limb fractures [25].

Plain lateral radiographs and axial CT cuts may miss lesions at the craniocervical junction, although flexion–extension views may discover an otherwise unnoticed luxation [20]. Accordingly, coronal and sagittal CT reconstructions constitute the neuroimaging method of choice [19, 25–27]. Findings of cervical spine CT include prevertebral soft tissue edema, C1–C2 subluxation, separation of the C1–C2 articular facets and increased occipitocervical distance [25–27]. Magnetic resonance (MR) imaging findings

Table 1Normal values used for evaluating our cases of severe cra-niocervical injury [19, 25–27, 39]

Anatomical structure	Upper limit of normal values
Retropharyngeal space	<7 mm
Retrotracheal space	<14 mm
Atlantodental interval	<5 mm
Lateral mass interval	<2.6 mm
Distance basion-odontoid tip	<12 mm
Interspinous C1-C2: C2-C3 ratio	≤2.5
Rule of thirds: Spinal canal, 1/3 dens	; 1/3 cord; 1/3 empty

comprise prevertebral, interspinous, or nuchal ligament swelling, C0–C1–C2 joint widening or fluid, possible epidural hematoma, and hyperintensity of the spinal cord [25–27]. Neurovascular injuries include lacerations of the pontomedullary junction, stretching or laceration of the midbrain, and contusion or laceration of the lower medulla and/or of the rostral spinal cord [25]. Subarachnoid, subdural, or epidural hemorrhages may also occur as well as cerebellar infarctions and spasm or dissection of the cervical carotid or vertebral arteries [25].

#### Management and outcome

Management starts at the accident scene and includes an adequate neck immobilization to avoid secondary spinal cord damage [28–37]. Ordinary resuscitation measures (breathing, blood pressure, and oxygenation), including orotracheal intubation if necessary, are given priority. Life-threatening lesions (pneumothorax, viscus rupture, etc.) are treated first. Only after patients' stabilization, the spinal lesion is addressed performing the appropriate neuroimaging studies.

General principles for the management of vertebral and spinal cord injuries comprise: (a) management of the osteoligamentous lesion and (b) management of the spinal cord injury when it exists. The basis of treatment for the osteoligamentous injury include: (a) identification of the type and level of the lesion looking for fractures or luxations and for signs of instability, (b) correction of spinal displacements and cord decompression if present, (c) fixation if there are signs of instability, (d) repeated follow-up studies for detecting late-appearing deformities or instabilities that would indicate the need for fixation [29, 33, 37, 38].

In cases of high cervical spinal cord involvement, there may be respiratory paralysis that often mandates intubation or tracheostomy. The efficacy of treatment with methylprednisolone in pediatric spinal cord lesions has not been fully investigated but its use is currently accepted because there is not a better alternative. Some conditions require emergency decompression, as are hematomas and bone or disk fragments within the spinal canal.

There is a general agreement in avoiding cranial traction in AOD as this maneuver could further increase the separation of craniocervical structures and intensify neural damage. In the case of craniocervical injuries, there are no universally accepted guidelines [33]. In general, a conservative attitude is acceptable when there is no instability or when the children's situation precludes surgical treatment [33]. However, even in the most severe instances, internal fixation may be indicated to allow for early rehabilitation once hemodynamic stability has been achieved. In cases with incomplete, or even with complete, spinal cord lesions, Fig. 1 Radiographic studies performed to patient 1. a Radiograph of the cervical spine showing increased atlantodental space (*arrow*); b CT showing this separation (*arrow*) and increased interspinous distance C1–C2 (*line*)



spinal fusion may prevent further neurological deterioration or result in significant neurological improvement [33, 37]. Ethical problems usually arise on managing children with apnea and quadriplegia who are ventilator dependent and who show evidence of medullary or high cervical cord transection by neuroimaging studies [38]. In our experience, surgery should be considered individually and after taking into account the attitude of the patients' parents. Accordingly, some authors advocate external immobilization with a hard collar or with a halo device even during prolonged periods of time. At present, most authors will advice internal fixation with wires, bone grafts, and, more recently with transarticular screws [34-36]. In AOD injuries, the occiput must be included in the craniocervical fixation [33, 35, 37]. A posterior fusion of the atlanto-axial segment is usually performed for unstable AAD injuries [28-37]. On performing an internal stabilization, it is advisable to consider the need of fixing the

Fig. 2 Neuroimaging studies corresponding to patient2. a Reformatted cervical CT showing enlarged atlantodental distance and widening of the C1–C2 interspinous dimension. b 3D-CT reconstruction clearly illustrating the widening of the C1–C2 joints. c MR depicting increased signal at the junction of the medulla with the cervical cord and an image of cord transection (*arrow*) occipital to C1–C2 by evaluating the ligamentous complex in MR studies. Again, there is no agreement on the use of postoperative external fixation with a halo device taking into account the fragility of the children's skull bones, for which some authors indicate that neck immobilization with a hard collar may usually be sufficient or even unnecessary [34].

Obviously, neurological outcome is closely related with the degree of the medullary and/or spinal cord injury, with coexisting cerebral lesions, and with the effects of initial hypoxia and arterial hypotension [38]. Good long-term structural results have been reported following internal fixation procedures for high cervical spine lesions [28, 29, 33, 37], although new methods that involve placement of screws are not devoid of complications such as vertebral artery injury [34, 35]. In Sun et al.'s series of children with occipitoatlanto-axial injuries, only 4 of 20 had spinal cord injuries, three of them were submitted to occipital–C2



fusions and the fourth died [39]. Of the remaining patients, treatment consisted of a craniothoracic orthesis (n=5), a collar (n=4), a halo device (n=1), and five received no treatment [39]. Neurologically, 13 patients were intact or asymptomatic, three showed developmental or cognitive delay, one had a hemiparesis and another one quadriparesis, one patient was lost to follow-up, and another one died [39]. All patients submitted to follow-up flexion–extension studies (n=11) showed good stability [39]. Undoubtedly, this series comprise a different population to the one that we are reporting.

## **Illustrative cases**

After permission from the Institutional Review Board, we surveyed the medical records pertaining to 20 children diagnosed with a craniocervical injury admitted to our hospital during the period 1997–2012. We excluded from the study patients with rotatory atlanto-axial subluxation (n=4), cases

Fig. 3 Neuroimaging findings of patient3. a Cranial CT showing a retroclival hematoma and subdural hemorrhage around the left cerebellar hemisphere. b Sagittal reformatted craniocervical CT exhibiting excessive separation of the atlantodental interval and of the interspinous distance. c 3D-CT reformatted views showing the typical findings of AAD. d MR illustrating abnormal signal at the bulbomedullary junction and disruption of spinal cord fibers, together with prevertebral edema and a fluidfilled cavity

of Grisel's syndrome (n=4), instances of congenital origin (Morquio and Down syndrome; n=3), one case of subluxation associated with a high cervical epidural hematoma, and three cases of high cervical spinal cord injury without radiographic abnormality.

*Case 1* This 8-year-old boy, hit by a car, was admitted in deep coma (Glasgow Coma Scale (GCS) score=3), apnea, and quadriplegia. A head CT scan showed diffuse brain edema and cervical radiographs and CT scan revealed a severe AAD (Fig. 1). In spite of all measures for hemodynamic stabilization, the child developed an acute renal insufficiency and died 3 days later.

*Case 2* This 4-year-old girl was a car occupant, appropriately restrained in a special seat, who was involved in a traffic accident. At admission in a nearby hospital where she was intubated, the child was apneic and had a GCS score of 6. At arrival to our center, she wore a cervical collar and was



found to be quadriplegic. A CT head scan showed smallscattered hemorrhagic lesions in the right frontal region and blood within the fourth ventricle. A cervical CT showed a severe C1-C2 distraction injury (Fig. 2a, b). A cervical MR demonstrated, in addition to AAD, decreased signal intensities extending from the medulla to C2, and an almost complete C1-C2 cord transection (Fig. 2c). Evoked potential showed a block in the conduction at a high cervical level with preservation of brain stem activity. The child also had a coagulopathy and bilateral pulmonary contusions. After hemodynamic stabilization, 12 days after the accident, she was given a posterior cervical fusion (Gallie technique). She was discharged to a national rehabilitation center after having been given a tracheostomy. After hospital discharge, no improvement in the girl's neurological condition was observed and she remained ventilator dependent. In the following 18 months, she had two further admissions due to respiratory problems and finally she was lost to follow-up.

*Case 3* This 10-month-old girl sustained a severe craniocervical traumatism when she was traveling correctly restrained in the back seat of an automobile that was involved in a frontal collision. The parents initiated basic resuscitation measures until the arrival of the emergency services that proceeded to advanced cardiopulmonary resuscitation, orotracheal intubation, administration of adrenaline till obtaining central pulse, and to cervical immobilization. At admission to the pediatric intensive care unit (PICU) of our hospital, a CT head scan was performed that showed a thin interhemispheric occipital and tentorial subdural hematoma, a thick retroclival hemorrhage with probable intraspinal

Fig. 4 Patient 4, craniocervical MR showing a contusion and edema at the bulbomedullary junction (*arrow*) and b a zone of cerebellar infarction, c CT-angiography demonstrating the interruption of flow in the segment V2 of the vertebral arteries

extension, and intraventricular bleeding with incipient hydrocephalus (Fig. 3a). An external ventricular drainage was inserted for intracranial pressure monitoring that was maintained for 3 days. Laboratory findings demonstrated hypoxemia, hypercarbia, and metabolic acidosis, together with severe anemia that necessitated a blood transfusion. A spine CT revealed an increased anterior atlanto-dens interval and separation of the C1–C2 spinous processes (Fig. 3b). There were no fractures and in the 3D reformatted images, a separation of >10 mm of the C1-C2 joints was appreciated (Fig. 3c). MR revealed hemorrhagic contusion and edema extending from the medulla oblongata to C4 and a line of probable cord transection at C2 together with an anterior fluid collection (Fig. 3d). Somatosensory evoked potentials showed a complete block in conduction at the bulbomedullary junction. In addition, there were posterior contusions of the left lung evidenced in the thoracic CT. During the child's stay in the PICU, she grimaced and opened her eyes but was apneic and had a complete flaccid quadriplegia. There was marked hemodynamic instability with persistent hypotension and bradicardia. On day 13 after admission, the girl presented marked oxygen desaturation and died.

*Case 4* A 13-year-old-boy was found unconscious while he was playing close to a freight elevator, probably following a fall from a height of 2 m. He had an initial GCS score of 6 and hypoxia. The boy showed marked facial edema and an extense bilateral subgaleal hematoma. The boy was intubated at hospital admission. CT and MR were performed (Fig. 4). MR showed AOD with posterior displacement of the atlas with a rotational component, there was also displacement and



contusion of the medulla and rostral cervical cord, together with edema/hematoma in the prevertebral space (Fig. 4a). There were also findings of bilateral cerebellar infarction (Fig. 4b). CT confirmed the AOD and showed a retroclival thick layer of subarachnoid hemorrhage that prompted an angio-CT with supraaortic vessels study that revealed a stop of the contrast at the segment V3 of the vertebral arteries (Fig. 4c). CT also showed a mandible fracture and bilateral pulmonary contusions. Three days after admission, the boy underwent a craniocervical fusion with a Vertex Max device that included screws in the lateral masses of C1 and traspedicular screws at C2 and C3. One week after the accident, a tracheostomy was performed. During his stay at the ICU, no motor activity or spontaneous breathing efforts were elicited when sedation was decreased to examine the patient's neurological evolution. Thirteen days after trauma, the boy died of an episode consistent with pneumonia and septic shock.

## Conclusions

Mild craniocervical traumatic injuries usually evolve with an intact neurological state and can be managed conservatively with only external immobilization. However, some craniocervical distraction injuries may produce severe damage to the brain stem and rostral cervical spinal cord. The rapid action of the emergency services has increased the survival of these patients that now arrive alive to hospital. The patients with severe neurological damage usually have unstable lesions due to significant ligamentous injuries and often require an internal fixation. Given that some cases with spinal cord injuries have been reported to improve after treatment and that these unfortunate patients need prompt rehabilitation, internal fixation is often regarded as a valid option as early as hemodynamic stability has been achieved. Serious ethical issues arise on considering the advisability of aggressive treatments taking into account the quality of life of the surviving children who are left ventilator dependent and quadriplegic and who show evidence of bulbomedullary or cord transection by neuroimaging studies. In our view, surgery must be considered in an individual manner; the attitude and beliefs of the parents being crucial in the process of decision taking.

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