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Pediatric spine and spinal cord injury after inflicted trauma

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Fortunately, children represent only a small fraction of the total number of patients affected by spine and spinal cord injury in the United States each year. The number of these injuries caused by intentional violence and child abuse is also small, but their impact to the patient, family, health care providers, and society in general is considerable. This article reviews the epidemiology, mechanisms of injury, developmental and radiographic anatomy, and neuropathology of inflicted spine and spinal cord injury in children. Although most inflicted spine and spinal cord injury occurs in the setting of child abuse, some cases are attributed to injury occurring outside the home by knives, firearms, or other physical assault. For the purposes of this article, the term *inflicted trauma* refers to intentional injury that occurs both inside and out of the home. A representative case of high cervical injury in an infant after violent shaking is included.

History, epidemiology, and demographics

An early description of spinal injury in children from the radiology literature unwittingly documented a group of infants subjected to physical abuse [1]. The author mistakenly attributed spinal and long bone fractures to "metaphyseal fragility," failing to recognize intentional spinal trauma in the absence of a corroborating history [2]. This lack of recognition is reflected in the paucity of information on intentional pediatric spinal trauma, which reveals very few series dealing directly with

* Corresponding author. E-mail address: rellen@chmc.org (R.G. Ellenbogen). this subject [3–5]. Most data are derived from articles [6–10] and texts [11] dealing with child abuse, which have included examples of spinal injury. In addition, there are numerous case reports documenting isolated cases of spinal injury from intentional trauma [12–22].

On the whole, pediatric spine and spinal cord injury (PSCI) are rare, the latter more so than the former. Fractures of the spine in children are uncommon and comprise less than 5% of all reported spinal injuries [9,23–25]. In the setting of child abuse, spine fractures have been noted in conjunction with other visceral injuries or long bone fractures, and are seen in less than 1% of nonaccidental trauma cases in most large series [10,26–31].

Several authors, however, believe that fractures in abused infants are much more common than previously reported [32–34]. One study showed that this figure may range as high as 3%, its author maintaining that a reason for the discrepancy may be related to a lack of consistency between various institutions in obtaining an adequate skeletal survey [6]. Kleinman's [11] review of the literature in the mid 1980s documented 41 cases of spinal fracture secondary to abuse, and pointed out over twice as many fractures were identified among these cases, attesting to the common multiplicity of such injuries.

The overall trend in spine and spinal cord injury is maintained in the setting of intentional trauma: spinal injury is more common than spinal cord injury, reflected in Swischuk's [5] original article on the subject, wherein six of the seven children presented with spine injury in the absence of a spinal cord injury. Given the small number of spinal cord injuries reported, it is difficult accurately to characterize the epidemiology of PSCI overall, let alone

in the setting of intentional trauma. The collection of population-based epidemiologic data and the publication of several large series over the past 30 years have brought about a consensus that the incidence of PSCI is between 1% and 10% of all spinal cord injuries that occur in the United States, resulting in 500 to 1000 cases of PSCI per year [1,25,31,36–38]. Among these series, the most common cause of traumatic PSCI has been motor vehicle accidents, primarily involving children as pedestrians or bicyclists struck by cars [31,35,37, 39,40]. In a small series, however, Haffner et al [41] showed that violence was equally responsible for PSCI in an urban population near Los Angeles, reflecting a disturbing trend also seen in the more urban subset of patients of one large series with injuries caused by gunshot wounds [40] and 13% of the patients in another series of 47 children [42].

Spinal cord injury caused by child abuse only constitutes a minor fraction of the cases in these large series [25,29,35,43] and in most series it is not mentioned [31,37,38,42]. Most series specifically addressing the issue of battered children show an equally rare incidence of spinal cord injury, most likely comprising less than 1% of the injuries that occur from this mechanism [6,7,10]. Several studies indicate that this is probably an underestimate [4,9]. In the series of Hadley et al [9] of 13 infants who sustained nonaccidental trauma, six of the children died as a result of head injury without evidence of direct cranial trauma, and five of these six underwent autopsy. An epidural or subdural hemorrhage was revealed at the cervicomedullary junction, and four of the patients had spinal contusions at upper cervical levels, leading the authors to conclude that hemorrhages and contusions of the high cervical spinal cord may contribute to morbidity and mortality [9]. Their study also questioned the dictum that cranial impact trauma is an essential component of the pathophysiology of the battered child syndrome [44].

Feldman et al [4] found similar although less dramatic results in the only prospective study done to date, wherein 12 children with central nervous system damage caused by intentional injury underwent MRI. Although the selection criteria differed from the series of Hadley et al [9], and none of the children had evidence of cervical cord injury on either conventional radiography or MRI, four of the five children who subsequently died underwent autopsy, revealing subdural or subarachnoid hemorrhage over the cervical cord. Feldman's [4]

study suggested that cervical spinal cord injury could be masked by concomitant head trauma, and stressed the need for vigilance in recognizing spinal cord dysfunction in the presence of coexistent head injury.

The literature underscores the fact that spine and spinal cord injury caused by child abuse may be underestimated in this age group. Unfortunately, spinal injury without a clear history of significant spinal trauma easily can be overlooked in an infant or child who is difficult to examine because of crying, limited verbal skills, or the inability to comply with a detailed neurologic examination [14]. In child abuse, confessions are rare, and the onus of responsibility for recognizing intentional trauma is placed on the examining physician. A detailed history and critical analysis relative to the physical signs and radiographic patterns is essential if abuse is to be recognized and adequately evaluated. Any incongruity between the history and postulated mechanism of injury should raise the suspicion of nonaccidental trauma.

Mechanisms and patterns of injury in intentional trauma

Understanding injury patterns suggestive of abuse can be of great benefit to the clinician in recognizing spinal injury after nonaccidental trauma. Unfortunately, given the limited numbers of cases, no unique pattern of injury becomes clear, and the general patterns of pediatric spine and spinal cord injury must be taken into account. Typically, younger children are susceptible to higher cervical injury related to their unique biomechanics and body habitus (discussed later). In the child who suffers intentional abuse, however, spine and spinal cord injury have been documented throughout the spinal axis, and vary according to the form and site of inflicted trauma.

Cervical injuries

In the cervical spine, the injuries seem to relate to whiplash-type injury mechanism, wherein the assailant grabs the shoulders and the child is forcibly shaken [9,22]. Alternatively, Piatt and Steinberg [20] proposed a different mechanism based on their reported case with facial petechiae and ecchymoses below the jaw. They hypothesized that the assailant held the child's head and whiplashed the body, resulting in a similar whiplash injury to the cervical spinal cord [20].

Regardless of the mechanism, the cervical spine of infants and small children is placed at increased risk of flexion, distraction, and rotational injuries because of horizontally oriented facet joints, incompletely formed uncovertebral joints, increased laxity of the surrounding ligaments, immature paraspinous musculature, and a higher head to body mass ratio [45]. Furthermore, the fulcrum for maximal flexion is at C2-3 in infants, and moves progressively caudal with age, resting at C5-6 after 12 years, as in adulthood [46].

Infants seem to be most susceptible to whiplash-shaking intentional trauma, with numerous case reports documenting craniocervical and high cervical injury of varying forms, including atlanto-occipital dissociation, atlantoaxial instability, hangman's fracture, subaxial instability, hematomyelia, epidural, subdural hemorrhage, and periadventitial extracranial vertebral artery hemorrhage and subsequent compression leading to stroke in the very young [16,19,21,22]. The following is a similar, representative case of an infant treated at the authors' institution.

Case report

This 24-day-old patient was a victim of nonaccidental trauma in the form of violent shaking. Sixteen hours after the incident, the mother noticed that the baby was cyanotic and unresponsive, and took the child to a local hospital where the infant was intubated and transferred to a trauma center. Several injuries including multiple rib fractures, a liver laceration, and a hip dislocation were noted. On neurologic examination, the infant had minimal tone in the lower extremities, minimal spontaneous movements with the upper extremities, and would only open eyes to voice. Initial cervical spine radiographs were interpreted as normal, but a CT scan of the head showed subdural and intraventricular hemorrhage.

The patient remained ventilator-dependent and hospitalized in an intensive care unit with prolonged sepsis. An MRI was obtained, which disclosed ligamentous injury at the occipitocervical junction, with atlantoaxial subluxation resulting in marked narrowing of the space available for the spinal cord (Fig. 1). CT imaging findings were consistent with disruption of the osseocartilaginous ring of C1, and sagittal atlantoaxial subluxation with rupture of the transverse ligament of the atlas (Fig. 2). An MRI of the brain demonstrated variable subacute subdural hemorrhage and evidence of hypoxic brain damage.

The patient was kept in a body cast with head immobilization for 2 months, at which time dynamic studies revealed widening of the atlantodental

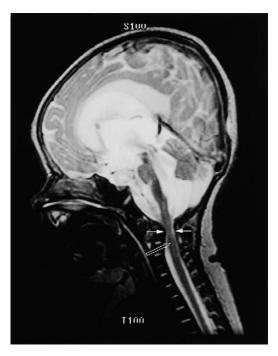


Fig. 1. Sagittal T2-weighted MR image demonstrates marked narrowing of the space available for the spinal cord at the occipitocervical junction (*solid arrows*). High signal within the lower brainstem and upper cervical spinal cord is consistent with resolving hematomyelia (*open arrow*). Significant brain atrophy is also noted.

interval unchanged in flexion and extension. The basion-dens interval remained unchanged on dynamic studies, which included gentle manual distraction. Her follow-up studies, 2 years after removal from the body cast, demonstrated persistent fixed widening of her atlantodental, and she underwent posterior surgical fusion at another institution. Her neurologic condition had stabilized and she was asymptomatic at the time of fusion. Although the patient has significant developmental delay and grade 4-5 strength in the right upper extremity, she is ambulating without assistance at latest follow-up (32 months).

This case highlights several important points about the history and mechanism of injury to an infant after intentional trauma. The infant was brought to medical attention after a prolonged period of listlessness and obtundation, and had a concomitant head injury, in addition to multiple other systemic injuries. The spine and spinal cord injuries were overlooked initially, with a substantial delay in diagnosis. The cognitive developmental delay seen in follow-up could be attributed both

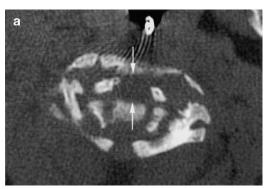




Fig. 2. (a) Axial CT scan through the ring of C1 shows disruption of the Osseo cartilaginous ring and marked widening of the atlantodental interval (*arrows*). (b) Sagittal CT reformat through the occipitocervical region reveals atlantoaxial subluxation and narrowing of the space available for the cord. Widening between the occiput and C1-2 complex is also demonstrated (*arrows*).

to the patient's head injury and hypoxic injury sustained during the period of cyanosis before the institution of treatment.

Thoracic and lumbar injury

Overall, the true incidence of thoracic and lumbar fractures in the general pediatric population is difficult to ascertain, but has been reported to represent 25% to 75% of all spinal fractures [24]. Given the limited numbers of cases in the literature, the incidence of thoracic and lumbar fractures after inflicted trauma, relative to cervical injuries, is even more difficult to characterize accurately. Nonetheless, some assert that most inflicted spinal injuries occur in the lower thoracic and upper lumbar vertebrae, at the apex of an acute kyphotic angle resulting from hyperflexion [3,5, 10,33].

In the thoracic and lumbar spine, the mechanism of injury is usually hyperflexion or hyperextension. Most infants with spinal injury have metaphyseal lesions that are characteristic of abuse, which occur because of the sudden acceleration and deceleration associated with hyperflexion and hyperextension during shaking [11]. The vertebral bodies are usually involved, most commonly in the form of varying degrees of compression fracture [11]. Anterior notching of the vertebral body and fracture-dislocations of the posterior elements are also common [5,10], but isolated posterior element fractures have also been recorded [11]. Carrion et al [12] documented a case of circumferential growth plate fracture at the thoracolumbar junction in two infants who suffered child abuse. Although no specific mechanism for the traumatic injuries was ever confessed by the parents, the authors hypothesized a mechanism of injury involving axial load, flexion, and rotation producing a circumferential fracture. In this previously unrecognized fracture pattern in child abuse, the centrum of the vertebral body separated from the disk and the growth centers and rotated into the spinal canal, resulting in complete paraplegia [12]. Such cases point to the unique nature of child abuse cases, because they can involve mechanisms that are unlikely to occur from any other injury type [12].

Pathology

In contrast to reports of survivors, injury patterns are well documented in autopsy series after nonaccidental trauma. The neuropathology of infants after atraumatic death is important to consider first, however, as a control. Towbin [22] observed epidural hemorrhage in five children who had died suddenly and unexpectedly, suggesting that this form of mechanical trauma could have contributed to their deaths. Subsequently, Harris and Adelson [47] reported 19 consecutive sudden and unexpected infant deaths, none of which were attributed to trauma, and documented varying degrees of epidural hemorrhage, greatest at the lower cervical region. This led them to conclude that the lower cervical epidural hemorrhages were atraumatic, the blood did not contribute to the deaths, and hemodynamic forces were responsible for the congestive phenomena and epidural bleeding.

The series by Hadley et al [9] was taken from 13 children who succumbed to child abuse. Here, the authors reported upper epidural cervical spine

hemorrhage in five of six infants, four of six also had subdural hemorrhage, and four of six had intramedullary contusions of the high cervical cord. Although the subdural cervical spine hemorrhages could have been attributed to subdural blood that tracked to the cervical spine by gravitational forces, the cervical cord contusions were an important and novel finding in abused infants.

The findings of Hadley et al [9] also have been supported at the microscopic level. Recent data from the neuropathology literature calls into question the widely held view that diffuse axonal injury is the main pathologic finding after nonaccidental trauma [48-50], and places the high cervical cord as the focal point in major morbidity and mortality after child abuse. Geddes et al [51,52] have recently published a large series of autopsy cases, using 37 infants aged 9 months or less who died from inflicted head injuries and 14 control infants who died from other causes, to show that diffuse hypoxic brain damage is the predominant histologic abnormality in cases of inflicted head injury. They found epidural cervical hemorrhage at the craniocervical junction in concert with focal axonal damage to the brainstem and spinal nerve roots in 11 cases of child abuse, but none in the controls, leading them to postulate that injury to the craniocervical junction could lead to apnea, hypoxic damage, and brain swelling. This, in turn, led to diffuse hypoxic brain injury that was previously believed to represent diffuse axonal injury caused by shear forces [52].

Prevention

The widespread proliferation of firearms in both urban and suburban populations in the United States among school-aged children, and the recent well-publicized mass shootings in suburban areas that have resulted in PSCI and spine injury cases, underscore the tragic rise in violent PSCI and the need for an urgent response [40,41]. Although a more recent study of the epidemiology of urban pediatric neurologic trauma only identified a small number of PSCI cases, the authors' conclusion on the prevention of neurologic trauma as a whole is germane to intentional spinal cord trauma prevention in children [8]. They found a greater relative reduction in injuries in the age and population groups targeted by an injury prevention program compared with untargeted controls. The authors concluded that there was a positive effect from targeting families at risk, providing novel education programs to parents regarding infant abuse and neglect, along with school-based programs to address conflict resolution [8].

Summary

Pediatric spine and spinal cord injury are rare sequelae of intentional trauma. They may easily be overlooked, however, and probably represent an underreported phenomenon. Recent autopsy data analyzed in conjunction with prior case series indicate that injury to the upper cervical spine and brainstem may significantly contribute to the major morbidity, mortality, and neuropathology in shaken infants.

The findings in the previous case report illustrate several important points regarding spine and spinal cord injury after intentional trauma. First, the very young are susceptible to severe, higher cervical injury of both spine and spinal cord. Second, spine and spinal cord injury were initially overlooked because of masked neurologic findings with the concomitant head injury and multiple other systemic injuries. Finally, the child's outcome with significant cognitive delay because of global brain injury in conjunction with the focal high cervical cord injury may support the hypothesis that hypoxic damage could have occurred secondary to brainstem and high cervical cord injury.

At the authors' institution, a detailed history and vigilant physical examination are stressed. When the mechanism of injury reported in the history is incongruous with the physical or initial radiographic findings and intentional trauma is suspected, a full skeletal survey, ophthalmologic evaluation, and social evaluation is undertaken. MRI and CT scanning are individualized according to the clinical assessment.

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