

# Pediatric Spinal Cord Injury without Radiographic Abnormality

*A Meta-analysis*

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Using a meta-analysis, we identified 392 published cases of patients recently diagnosed with spinal cord injuries without radiographic abnormalities (SCIWORA) and studied the epidemiologic, pathophysiologic, clinical, and radiologic data. To describe those at risk for this uncommon syndrome, mainly pediatric patients (90% of the cases) who sustain serious trauma in car accidents, serious falls, sports injuries, or child abuse, we analyzed the reported information in the literature. Magnetic resonance imaging scans may indicate neural (hemorrhages, edema, or both) or extraneural (disc protrusions, extradural hematomas) injuries, and the location and type of the injury. Every patient having magnetic resonance imaging scans had either intraneural or extraneural injury. However, followup magnetic resonance imaging scans are necessary because evidence of the injury might not appear immediately. Late and recurrent spinal cord injuries without radiographic abnormalities are reported. External immobilization for 12 weeks helps patients who are moderately injured and helps prevent recurrence of these types of injuries. Approximately 44% of the patients in our study did not recover whereas complete recovery occurred in 39% of the population. The prognosis can be improved if the syndrome is diagnosed early, so we recommend considering spinal cord injuries without radiographic abnormalities as a possible diagnosis for any child who has a mechanism of injury that suggests trauma to the spine.

**Level of Evidence: Therapeutic study, Level III-3 (case-control study). See the Guidelines for Authors for a complete description of levels of evidence.**

Spinal cord injury without radiographic abnormality (SCIWORA) describes an acute spinal cord injury including trauma to nerve roots that may result in some degree of sensory or motor deficit, or both,<sup>13</sup> without evidence of vertebral fracture or malalignment on plain radiographs and CT scans.<sup>9,11,25</sup> The concept of spinal cord injury without radiographic abnormality was proposed by Lloyd<sup>19</sup> in 1907, but its acronym, SCIWORA, was coined by Pang and Wilberger in 1982.<sup>25</sup>

Although relatively rare, the SCIWORA syndrome presents a broad clinical spectrum, and because the injury is not identifiable in routine radiographs or CT scans which is self-evident from the syndrome's name, a diagnosis can be difficult if the potential for its occurrence is not considered. Because SCIWORA most often is caused by serious trauma (motor vehicle accident, significant fall, sports injury, child abuse) and can result in serious and irreversible consequences for the patient, the most important challenge to the clinician is to recognize this syndrome as quickly as possible so that treatment can be instituted early. Adding magnetic resonance imaging (MRI) scans to the diagnostic workup may show neural and extraneural injuries and indicates that the clinician is alert to a potential neural or spinal cord injury. Our meta-analysis looked for ways to understand the characteristics of the patient population to determine when a child is at risk for SCIWORA, and to describe the diagnosis of this syndrome.

After the diagnosis of SCIWORA was tentatively made (from the evidence of neurologic examination, appropriate MRI scans, negative radiographs), various treatment protocols were attempted at various institutions. We examined them to see if there were any clues to optimum management of SCIWORA. Some clinicians have tried external immobilization of the patient for varying times. Others

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combine immobilization with the use of steroids. After the first scans, additional MRIs may provide information that confirms the diagnosis.

Will a meta-analysis of a patient population already diagnosed with SCIWORA syndrome, as described in the literature, help clinicians make a timely and correct diagnosis? Will a review of the treatment protocol at various institutions help a clinician decide such questions as whether to immobilize a patient and for how long? Are there any clues to the optimum management of treating SCIWORA from a synthesis of the information available in the current literature?

## MATERIALS AND METHODS

To identify published data about SCIWORA, we reviewed the Medline database from January 1980 through May 2002 using the key words "SCIWORA" (all fields) and "acute spinal cord injury" or "acute spinal cord trauma" (title only). Three hundred ninety-eight articles were found.

To be included in the study, authors had to discuss SCIWORA in general or report patients with posttraumatic neurologic deficit and no visible injury seen on plain radiographs. Articles were excluded if they included patients with (1) radiographic or CT evidence of vertebral column injury or (2) posttraumatic neurologic deficit without radiographic or CT information. The references cited in each eligible article were analyzed according to the same inclusion and exclusion criteria. Eighty articles satisfied our criteria and yielded 392 cases of SCIWORA syndrome. The information available in each case was categorized in terms of epidemiology, pathophysiology, diagnosis, and prognosis.

From the literature, information was obtained characterizing the population of patients with SCIWORA for age, gender, location of spinal injury, etiology, mechanism of injury, examination on presentation, and workup, including plain radiographs and MRI studies. A separate subpopulation of patients with recurrent SCIWORA was studied to better understand this condition. Finally, the treatment protocols for all patients with SCIWORA were reviewed to determine optimal treatment for this population.

## RESULTS

There were 353 pediatric patients (90%) and 39 adult patients (10%). Of the 353 identified pediatric patients, the exact age was available for 132 patients (37%); the rest were defined as 18 years or younger. In the results that follow, for each parameter discussed, the denominator represents the total number of patients for whom information was provided about that parameter. The average age at injury for the 132 patients with their exact age available was 7.4 years (range, 0–18 years). Gender information was

available for 152 pediatric patients; of these patients, 90 were boys (59%) and 62 were girls (41%).

Data on the location, cause, and mechanism of the injury (when known) were classified. Information on the specific location of the spinal injury was available for 92 of the 353 pediatric patients. Of these patients, the spinal injury was located in the cervical spine in 68 patients (74%) and in the thoracic spine in 24 patients (26%). The etiology was identified in 115 (44%) of the pediatric patients and was a motor vehicle accident in 52 patients (45%); a fall from a height in 31 patients (27%); a sports-related injury in 15 patients (13%); birth trauma in 14 patients (12%); and child abuse in three patients (3%). The specific mechanism of injury for the SCIWORA syndrome was available for 105 patients and was flexion in 57 patients (54%); distraction in 22 patients (21%); extension in 19 patients (18%); and crush in seven patients (7%).

The presentation of patients in these case studies showed a wide clinical spectrum. The studies indicated five different clinical presentations in 133 children: partial cord syndrome, 73 patients (55%); complete cord syndrome, 36 patients (27%); central cord syndrome, 13 patients (10%); Brown-Séquard syndrome, seven patients (5%); and anterior cord syndrome, four patients (3%). However, the initial clinical condition was available only for 20 pediatric patients. Sixteen of them were hemodynamically unstable (80%), requiring admission to an intensive care unit. No child was reported to be neurologically unstable. Twenty-four of 88 patients (27%) presented with delayed symptoms, and 15 of 22 patients (68%) were identified as having premonitory symptoms.

When initial MRI scans were obtained, information regarding specific areas of injury (intraneural or extraneural) was available for review. In no patients was the MRI normal for intraneural and extraneural tissues. Data from MRI scans were available for 13 pediatric patients and showed hemorrhage around the spinal cord in six patients, edema in three patients, and hemorrhage and edema in two patients. Two patients had no MRI evidence of intraneural injury. Extraneural findings in these same 13 patients included disc herniation in two patients, disruption of the posterior longitudinal ligament in one patient, disruption of the anterior longitudinal ligament in one patient, and disruption of the interspinous ligament in one patient. The extraneural tissues were normal on MRI scans in four patients. Data for the four remaining patients did not include radiographic information about the extraneural tissues.

Treatment consisting of rigid immobilization seems more effective if of longer duration (12 weeks rather than 8 weeks), although approximately 44% of patients showed no recovery after treatment. Treatment protocol and outcomes were documented for 98 pediatric patients. One

patient who was treated with a combination of corticosteroids, mannitol, and hyperbaric oxygen therapy, recovered partially. Another patient who was treated by a combination of corticosteroids, mannitol, and external rigid immobilization for 12 weeks, recovered partially. Three patients did not receive any treatment, one of them died, and the other two had no neurologic recovery. The other 93 patients were treated with external rigid immobilization and no reported medication for 8 weeks (41 patients) or 12 weeks (52 patients). In the patients treated for 8 weeks, two patients died (5%), 18 had no recovery (44%), seven recovered partially (17%), and 14 recovered completely (34%). In the patients treated for 12 weeks, one patient died (2%), 23 had no recovery (44%), six recovered partially (12%), and 22 (42%) recovered completely (Table 1).

Of the 41 patients immobilized for 8 weeks, seven (17.1%) had recurrent SCIWORA develop, whereas no patient immobilized for 12 weeks had recurrent SCIWORA develop. The decrease in SCIWORA in the group of patients who were immobilized for 12 weeks compared with being immobilized for 8 weeks was statistically significant (Fisher's test,  $p = 0.01$ ).

Recurrent SCIWORA, defined by the onset of a second SCIWORA several days to several weeks after the initial SCIWORA,<sup>24,26,31</sup> was diagnosed in 16 of 97 pediatric patients (17%).

The neurologic deficit or recovery after treatment was available for 109 patients. Complete recovery occurred in 36 patients (33%), partial recovery occurred in 16 patients (15%), no recovery occurred in 53 patients (49%), and four patients died (4%).

## DISCUSSION

Although this meta-analysis was limited by the small proportion of patients with complete data available, sufficient patients remained in the subgroups to provide meaningful information. Another limitation was the varying degree of treatment protocols at various institutions. For example, medication protocols were variable, therefore the effect of steroids or other medications could not be determined.

**TABLE 1. Patients (n = 93) Treated with Immobilization Only (No Medications)**

Outcome	Immobilized 8 Weeks (41 patients)	Immobilized 12 Weeks (52 patients)
Died	2 (5%)	1 (2%)
No recovery	18 (44%)	23 (44%)
Partial recovery	7 (17%)	6 (12%)
Complete recovery	14 (34%)	22 (42%)

Some authors have reported that SCIWORA occurs almost exclusively in children,<sup>7,23</sup> but some authors have reported some occurrences among adults (10% of cases).<sup>11,15</sup> Therefore, it may be more accurate to state that SCIWORA syndrome can occur in patients of all ages, but it occurs predominantly in the pediatric population.<sup>14–16,24</sup>

In the United States, the reported incidence of all spinal cord injuries in the pediatric population is 18.1 spinal cord injuries per year per million children; representing approximately 1300 new cases per year.<sup>14</sup> However, determining the exact incidence of spinal cord injuries in the pediatric population is difficult because, in some reports, adult and pediatric patients are combined in a general population. The incidence of children's cases in this overall group varies from 0.6–59.6%.<sup>8,13,23</sup> One study that does define the incidence of all spinal cord injuries in the pediatric population is the 1980 epidemiologic analysis by Kewalramani et al.<sup>13</sup> Although somewhat dated, the analysis offers the advantage of estimating the number of pediatric spinal cord injuries in relation to the overall population of children in a geographic area.

An accurate estimate of SCIWORA in the pediatric population ranges from 19–34% of all spinal cord injuries; although earlier reported estimates ranged from 1.3–67%.<sup>11,22,25,29</sup> This earlier wide range can be attributed to the following factors. First, some percentages may be biased because they are not based on careful analysis of comparable cases or on results of original investigations.<sup>11,12,21</sup> Second, some authors calculated the prevalence of SCIWORA in children with spinal cord or vertebral column injury, or both.<sup>5,29</sup> For example, Dickman et al<sup>5</sup> reported 24 cases of SCIWORA syndrome in 159 patients with spinal (or vertebral) column injuries (13.4%), and Turgut et al<sup>29</sup> reported 11 cases in 82 children (16%). However, if the numbers of vertebral column injuries without spinal cord injury which were included in these studies are deleted, the prevalence of SCIWORA becomes greater than 16%. Third, another study included only the cervical spine.<sup>2</sup> Finally, certain series data derived from rehabilitation facilities are not representative of the true incidence of injury but rather, represent children who received extended medical care.<sup>1,22</sup> Therefore, a more accurate incidence of SCIWORA in pediatric spinal cord injuries ranges from 19–34%.<sup>14,23,27</sup>

Spinal cord injury without radiographic abnormality syndrome is a result of serious trauma, and the diagnosis may be complicated by other serious injuries. The most common cause of SCIWORA is motor vehicle accidents.<sup>1,3,12,13</sup> Affected children who are younger than 8 years are usually motor vehicle passengers, unrestrained (or improperly restrained) by car seats or seat belts, or pedestrians struck by vehicles.<sup>20,21</sup> Affected children who are 8 years or older usually are struck while cycling.<sup>21</sup>

Other serious pediatric traumas are caused by child abuse or sports. A special case for infants is the rigors of childbirth.

Three anatomic characteristics have been proposed to explain the much higher incidence of SCIWORA in children than in adults, producing a neural injury rather than a fracture. First, in children, the facet joints (and particularly those in the upper cervical spine) have a more horizontal orientation,<sup>7,11,14,16,23</sup> allowing increased translational motion in an AP plane.<sup>4,21,22,24</sup> Second, children have anterior wedging of the superior aspects of the vertebral bodies.<sup>11</sup> Third, pediatric ligaments and joint capsules have greater elasticity.<sup>6,24,25,30,31</sup> These features allow for excessive intersegmental movement and transient soft disc protrusion, resulting in neural injury without fracture of the vertebrae or frank rupture of the ligaments in the spinal column.<sup>4,9,15,26</sup>

The location of injury in SCIWORA commonly involves the cervical cord secondary to forced excessive spinal movement,<sup>28,31</sup> particularly the upper cervical cord in children younger than 3 years.<sup>12,18,24,25</sup> Upper cervical SCIWORA has been reported to have more severe neurologic consequences than lower cervical SCIWORA.<sup>24</sup> Thoracic SCIWORA is less common because the splinting effect of the rib cage prevents the thoracic spine from forced flexion or extension.<sup>24,31</sup> Spinal cord injury without radiographic abnormality in the lumbar spine is very rare<sup>28</sup> and, to our knowledge, there are no reports of lumbar SCIWORA in children; probably because of associated lethal injuries such as aorta rupture.

Some reports indicate that some patients with SCIWORA have a latent period before neurologic deficits occur.<sup>10,18,31</sup> In retrospect, a history of premonitory neurologic symptoms often is elicited.<sup>16,24,25</sup> This delay in diagnosis may be secondary to the development of spinal cord ischemia or spinal instability.<sup>24</sup> Because a child often is unresponsive to commands or is medically unstable, an accurate neurologic examination may be difficult.

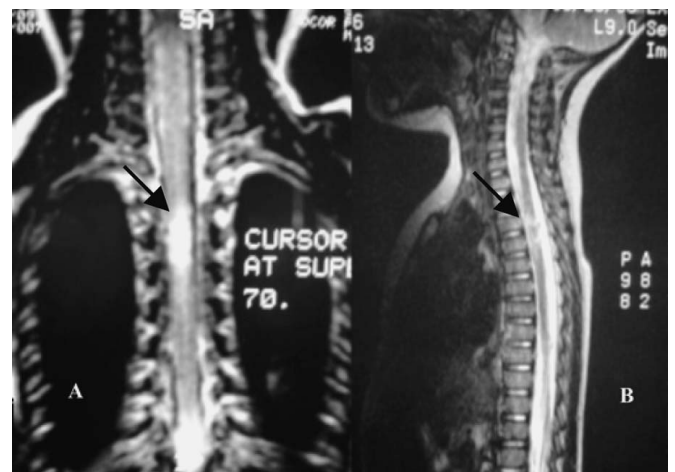
When neurologic deficit is diagnosed clinically but neither plain radiographs nor CT scans show any abnormality, MRI scans become the only way to show neural and extraneural injuries. Some clinicians recommend dynamic radiographs to assess potential spinal instability,<sup>4</sup> but acute dynamic radiographs may not show the subtle amount of subluxation because the paraspinous musculature often splints the spine and dynamic radiographs might cause additional injury to the spinal cord in an unresponsive patient.

For delineating spinal cord and soft tissue abnormalities,<sup>21</sup> MRI scans are superior to CT<sup>9,11</sup> in detecting neural injuries such as edema and hemorrhage,<sup>9,31</sup> and extraneural injuries such as traumatic disc protrusions, compressive extradural hematomas, and ligamentous injuries.<sup>15</sup> In ad-

dition, abnormalities observed on MRI scans correlate closely with the type of the neurologic injury.<sup>10,11,17</sup> Intramedullary hemorrhage seen on MRI scans is predictive for complete cord injury,<sup>9</sup> and the level of the MRI abnormality corresponds with the level of neurologic dysfunction.<sup>9</sup> Therefore, to assess injuries and their evolution, some clinicians recommended routinely obtaining initial and followup MRI scans in all pediatric patients with suspected SCIWORA.<sup>6,9</sup> Our meta-analysis also supports a trauma treatment protocol including appropriate MRI scans after serious trauma, with repeat studies at appropriate intervals, as the most useful diagnostic tool.

Followup MRI scanning has become important because SCIWORA can recur.<sup>16,21,23,26</sup> Resolution of the changes seen on MRI scans predicts a return to normal neurologic function, whereas persistence of spinal cord injury lessens the chances of a good outcome.<sup>6,8,31</sup> Recurrent SCIWORA is secondary to early spine mobilization from absent or inadequate treatment (Fig 1) and often results in more severe deficits than the initial syndrome.<sup>21,31</sup>

Recommended treatment is essentially nonoperative, including corticosteroids<sup>10,16</sup> and permanent (night and day) rigid external immobilization<sup>16,18,23</sup> for 12 weeks rather than for 8 weeks.<sup>9,24,26</sup> Although longer immobilization is counterintuitive given the fact that most bone and ligament injuries heal more quickly in children, our analysis supports prolonged immobilization. Traction and lami-



**Fig 1A–B.** (A) An MRI scan shows a contusion of the spinal cord at T3–T4 (arrow) without edema, hemorrhage, or extraneural injury. The patient, a 2-year-old girl, was noted to be neurologically intact on initial examination in the emergency room and had normal plain films of the spine. (B) This MRI was obtained after subsequent deterioration in neurologic status. This case illustrates that SCIWORA can manifest symptoms well after initial presentation and must be considered in the differential diagnosis of neurologically intact children who have a significant trauma history.

nectomy are not indicated.<sup>30</sup> Surgery may be useful only for patients with spinal cord compression or spine instability secondary to extraneural injury.

An initially severe neural injury is associated with a poor prognosis,<sup>21,25,31</sup> whereas an initially mild to moderate injury often is followed by a good recovery.<sup>21,31</sup> Prognosis depends on initial clinical instability, neurologic features, the age of the child, and MRI findings. Children younger than 8 years experience greater neurologic trauma<sup>4,5,16</sup> and have a worse prognosis than older children.<sup>7,21,23</sup> Findings seen on MRI scans indicative of cord transection and major hemorrhage are correlated with poor outcome,<sup>9</sup> minor hemorrhage or edema only is associated with moderate to good recovery,<sup>5,8,16,29</sup> and the absence of any abnormal cord signal suggests the patient will make a complete recovery.<sup>15,17</sup> Because SCIWORA has a relatively poor prognosis, but one that can be improved if diagnosed early, we recommend including SCIWORA in pediatric spine injury protocols.

If a neurologic examination of a child with a trauma history is abnormal, or if it is normal but with initial transient symptoms (for example, transient numbness), the child should be treated as if he or she has a spinal cord injury and should be immobilized appropriately. If plain radiographs and CT scans do not show any vertebral damage, MRI scans should be obtained to rule out SCIWORA. However, if the child is unconscious after injury, rigid immobilization should be applied, and plain radiographs and CT scans should be obtained. If these studies are normal, MRI should be obtained.

Because MRI may identify SCIWORA, and because the more definitive dynamic radiographs might aggravate an existing spinal cord injury, the best treatment algorithm is to maintain a patient's immobilization until consciousness improves to the extent that neurologic assessment and assessment of back pain can be obtained.

Once SCIWORA is diagnosed, maintaining immobilization, even after the return of normal neurologic function, for a full 12-week course may prevent additional damage to the cord. Prolonged immobilization also seems to reduce the chance of recurrent SCIWORA.

Because of the implications of SCIWORA and the time and expense involved in treatment and prevention, a multicenter prospective study with detailed case reporting and a standardized treatment protocol would be useful to substantiate the findings of this meta-analysis.

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