

Management and Mid-Term Outcome After "Real SCIWORA" in Children and Adolescents

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Abstract

Study Design: Retrospective analysis.

Objective: The SCIWORA Syndrome (Spinal Cord Injury Without Radiographic Abnormalities) is a rare but potentially severe injury with a peak in childhood and adolescence. With a better understanding of injury patterns and advances in MRI, there is ongoing discussion regarding the "Real SCIWORA" syndrome, a clinical picture of neurologic deficits on clinical examination but absence of radiographic pathologies even on MRI. The purpose of this study was to evaluate mid-term clinical outcome and the psychological impact of the "Real SCIWORA."

Methods: In this retrospective analysis, we evaluated 32 patients treated for "Real SCIWORA" between 2007-2019. Inclusion criteria were: neurologic deficit after trauma, no other cerebral or skeletal injury and a lack of pathological findings in spinal MRI. All patients were followed until complete recovery from initial symptoms. 25/32 patients were re-evaluated after 6.9 years (1-14 years) using the Oswestry Disability Index, the Frankel Score, the EQ-5D score, and the Breslau Short Screening Scale for PTSD.

Results: Initial neurologic presentation ranged from Frankel Grade A-D. All patients recovered neurologically during 1-13 days to a Frankel Grade E. The analysis of HR-QoL revealed no difference between the cohort of SCIWORA patients and the German population norm, Oswestry Disability Index showed only minimal disabilities. 4/25 patients showed signs of PTSD.

Conclusions: The "Real SCIWORA" syndrome is a diagnosis per exclusionem requiring a full spinal MRI to ensure exclusion of structural and potentially serious reasons of the neurologic impairment. Further clinical re-evaluation, psychological support seems to be essential.

Level of Evidence: IV—retrospective study.

Keywords

pediatric and adolescent trauma, management of severe trauma, SCIWORA, patient-reported outcome

In 1982 Pang and Wilberger defined Spinal cord injury without radiographic abnormality or "SCIWORA" as "children with objective signs of myelopathy as a result of trauma, whose plain films of the spine, tomography, and occasionally myelography carried out at the time of admission showed no evidence of skeletal injury or subluxation."¹ They recommended the use of every radiographic diagnostic known at that time including tomography and flexion-extension radiographs to detect instability. They reported that patients with severe neurology

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Creative Commons Non Commercial No Derivs CC BY-NC-ND: This article is distributed under the terms of the Creative Commons Attribution-Non Commercial-NoDerivs 4.0 License (https://creativecommons.org/licenses/by-nc-nd/4.0/) which permits non-commercial use, reproduction and distribution of the work as published without adaptation or alteration, without further permission provided the original work is attributed as specified on the SAGE and Open Access pages (https://us.sagepub.com/en-us/nam/open-access-at-sage). do not recover.¹ But with advances in radiology this definition seems dated. During an era when radiological examination of the spinal cord was limited, it seems likely, that the patients treated for SCIWORA included patients with severely injured spinal cords. Our understanding of SCIWORA has evolved as the Magnetic Resonance Imaging (MRI) has improved and become widely available.^{2,3} MRIs enable clinicians to evaluate the intramedullary state of the spinal cord in detail and helps to identify diseases that may require surgical intervention. Also it can distinguish between spinal cord lesions, disc herniations, edema, hematoma, contusions, ischemic injuries and the "Real SCIWORA"—the absence of any spinal abnormality.⁴

Nonetheless, there are few studies that utilize MRI to rule out more specific diagnoses and use the SCIWORA syndrome as an umbrella term for unspecific injuries. The "Real SCIWORA" syndrome without any radiographic effect on MRI differs from the previously published imprecise definition of SCIWORA syndrome.

The purpose of this study was to evaluate children and adolescent patients who were treated for neurological deficit after trauma, without any pathological finding in spinal MRI to show the mid-term outcome regarding neurological disabilities as well as psychological impact of neurologic impairment.

Methods

The study was approved by the Ethics Committee of the University of Regensburg (Institutional Review Board Number 20-1928-101). Informed consent was obtained by all participants. We conducted a retrospective study on "Real SCIWORA" patients between 2007 and 2019 who were admitted to our level I trauma center for traumatic spinal injury.

Inclusion criteria were:

- Post-traumatic sensory-motoric deficit of the upper or lower extremity
- Absence of any sign of post-traumatic effect on a highresolution MRI scan of the complete spine

Exclusion criteria were:

- Any post-traumatic alteration of the signal within the MRI scan
- Any concomitant injury that might potentially cause a neurologic symptomatic (e.g. traumatic brain injury, fracture or laceration that might go along with a peripheral nerve injury)

The evaluation included all available X-rays, Computed tomography (CT) scans, and a full spinal MRI-scan of every patient. A radiologist and a senior trauma surgeon carried out evaluation. The diagnosis of spinal cord injury was established by a specialist (neurosurgeon or neurologist). All patients were followed until complete recovery of neurologic symptoms. After 6.9 years (1-14 years) we re-evaluated all available patients for an assessment of clinical, functional and psychological mid-term outcome. The Oswestry Disability Index was applied to quantify disease-specific functional outcome due to back pain. This questionnaire contains: intensity of pain, lifting, ability to care for oneself, ability to walk, ability to sit, sexual function, ability to stand, social life, sleep quality, and ability to travel.⁵

The Frankel Score was used to assess spinal cord function. It is a 5 grade scale to assess the extent of the neurological/functional deficit into 5 grades: The functional deficit is classified as complete (grade A), complete motor loss (grade B), incomplete motor loss without practical use (grade C), incomplete motor loss, able to ambulate with or without walking aids (grade D), or no neurological deficit/complete recovery (grade E).⁶

For evaluation of global health-related quality of life (HR-QoL), we used the EQ-5D-3 L score. The EQ-5D is a global quality of life questionnaire consisting of a 5-item assessment of the health status regarding mobility, self-care, usual activities, pain/discomfort, and anxiety/depression. The second part of the EQ 5-D consists of a 100 mm global health visual analogue scale (EQ-VAS) ranging from 0-100 points concerning the patient's assessment of their current global health status. The EQ-5D has been adapted to German and has been validated for a number of health compromising conditions.^{7,8}

For evaluation of possible posttraumatic stress disorder (PTSD), the Breslau Short Screening Scale (BSSS) for PTSD was applied. It contains a seven-symptom screening scale and assesses if the patient: avoids places and people; lost interest in activities; felt isolated; found it hard to have affection for others; had a sense of foreshortened future; had sleep difficulties; or became easily startled. It is well proven to be an efficient way to screen for PTSD.^{9,10}

Statistics

Statistical analysis was performed using the software package SPSS (Version 24, SPSS Inc, Chicago, Illinois). Level of significance was defined at p < 0.05 for all tests. Descriptive data are given as frequencies (n) and percentage (%) for categorical variables, means (m) and standard deviations (\pm) for continuous and normal distributed variables. Normal distribution was assessed by Shapiro-Wilk-Test.

Results

32 patients met the inclusion criteria of traumatic spinal injury which led to neurologic deficits without any other cerebral or skeletal injuries, who's MRI scans revealed no pathologic findings. The patients were between 5 and 21 years old (mean: 14.5 years). There were 11 (34.4%) female and 21 (65.6%) male patients. 11 patients were admitted to the intensive care unit (ICU) (34.4%) with a mean stay in the ICU of 1.3 days (1-3 days). The Glasgow Coma Scale (GCS) was 15 in all patients and the ASA physical status was I. The mean hospital stay was 2.75 days (1-13 days). No patient required surgical intervention. Most patients fell from height (43.8%). 25% of the patients were injured in a sporting accident, 18.8% had a motor vehicle accident and 12.5% of the patients got injured

Table 1. Baseline Characteristics of the Study Popula	tion.
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	SCIWO n	RA patients = 32
Age (in years)	14.5	(5/21)
Gender		
Female	11	(34.4%)
Male	21	(65.6%)
Intensive care unit		
No	21	(65.6%)
yes	11	(34.4%)
Intensive care unit stay (in days)	1.3	(1-3)
GCS	15	(14/15)
ASA physical status ⁴	1.0	(1.0/1.0)
In hospital stay (in days)	2.75	(1/13)
surgery yes	0	(0%)
no	32	(100%)
Mechanism of injury		
motor vehicle accident	6	(18.8%)
fall from height	14	(43.8%)
sporting accident	8	(25.0%)
playing	4	(12.5%)

Data show number of patients for categorical variables; ASA physical status (American Society of Anaesthesiologists Score); GCS (Glasgow Coma Scale).

while playing. Table 1 presents the baseline characteristics in 32 patients.

The initial clinical presentation of neurology was a complete motor and sensory loss (Frankel Grade A) in 6 cases. 3 patients showed a complete motor loss (Frankel Grade B), 12 patients presented an incomplete motor loss without practical use (Frankel Grade C) and 11 patients showed incomplete motor loss but were able to ambulate with or without walking aids (Frankel Grade D).

After 1-13 days, all patients showed complete neurologic recovery (Frankel Grade E). Table 2 presents the initial clinical presentation of Real SCIWORA patients.

After 6.9 years (1-14 years), 25/32 patients were available for assessment of mid- to long-term clinical and functional outcome. The analysis of HR-QoL revealed no difference between the cohort of SCIWORA patients and the German population norms¹¹ regarding both EQ-5D VAS and EQ-5D Index Value. The functional analysis using ODI showed only minimal disabilities in day to day life.

The BSSS revealed that 4 out of 25 patients showed signs of a relevant PTSD (Table 3).

Discussion

This is the largest series of patients evaluating mid- to longterm outcome after "Real SCIWORA" syndrome with complete absence of radiographic signs of trauma even in high-resolution MRI. We found excellent clinical and functional results with a full neurologic recovery in all patients. However, a high number of 4/25 patients suffered from a relevant PTSD after "Real SCOWORA". **Table 2.** Clinical Presentation of Real SCIWORA in the Study

 Population During Hospital Stay.

	$\begin{array}{c} \text{SCIWORA} \\ \text{patients} \\ n = 32 \end{array}$
Initial clinical presentation:	
Frankel Grade:	
A: Complete motor and sensory loss	6
B: Complete motor loss	3
C: Incomplete motor loss without practical use	12
D: Incomplete motor loss, able to ambulate with or without walking aids	11
E: free of neurological symptoms	-
Time to complete recovery in days:	3,9 d (I-I3d)
Clinical presentation after recovery:	
Frankel Grade:	
A: Complete motor and sensory loss	-
B: Complete motor loss	-
C: Incomplete motor loss without practical use	-
D: Incomplete motor loss, able to ambulate with or without walking aids	-
E: free of neurological symptoms	32

Initial clinical presentation of neurological symptoms in the study population and their neurologic status after full recovery using the Frankel Grading and time to complete recovery in all patients in days.

The incidence of SCIWORA described in literature shows a wide range between 3.3% and 37%, depending on patients' age and the use of MRI.^{12,13} The prevalence may be overestimated due to mis-classification and lack of MRI. The term "SCIWORA" is often inappropriately used as an umbrella term for clinical findings that are difficult to diagnose such as spinal cord concussion, brachial plexus, nerve root, or peripheral nerve injuries.¹³ Even though many articles deal with the topic of SCIWORA there is no consensus regarding the definition, demographics, etiology, treatment or prognosis.

SCIWORA is more commonly seen in the children and adolescents than in the older population, an observation we also made in our collective.^{14,15} Since SCIWORA often occurs as a result of hyperextension forces (e.g., cervical acceleration causing whiplash injuries in car accidents) or from a direct impact to the face, main causes of injury in literature for young children are road accidents, falls and child abuse. In adolescent years sporting accidents are getting more common. These observations are supported by our data as well as by reports about evolvement of injury mechanisms of children and adolescents in general.^{16,17,13,14}

Initial clinical assessment and anamnesis are more difficult with young patients with numerous factors (anxiety of patients and parents, lack of pediatric and adolescent clinical experience in emergency room, unconscious patient) complicating the first evaluation.¹⁸ Therefore initial management begins with spinal immobilization and clinical evaluation.

The initial neurological presentation is multifaceted, ranging from paresthesia, motor weakness, sensory loss, pathological reflexes, loss of rectal tone, incontinence and paralysis to

Table 3. Mid-Term to Long-term Clinical Results After Real

 SCIWORA in the Study Population.

	SCIWORA Patients n = 25
Health related Quality of Life:	98.48
EQ-5D VAS	0,957
EQ-5D Index Value	
Oswestry Disability Index	6,04% (0-20%)
Posttraumatic stress disorder (Breslau short screening scale)	4/25

Mid- to long-term outcome of 25 Real SCIWORA patients using the EQ-5D fort the evaluation of health-related Quality of Life, the Oswestry Disability Index and the Breslau short screening scale for an evaluation of posttraumatic stress disorder.

partial or complete cord syndrome. (16) Knox et al. described additional injuries in up to 87% of the cases of SCIWORA. Young patients show the highest rate of additional injuries, particularly injuries to the head, face, and extremities.¹⁹ We ruled out any other traumatic cause for peripheral neurologic symptoms by excluding all patients who were otherwise injured for an outcome evaluation of only Real-SCIWORA patients. The treatment of "Real SCIWORA" is conservatively. In literature, hard collar bracing for 12 weeks is postulated as standard of care.⁴ Pang et al. recommended bracing of patients with persistent neurologic deficits but normal MRI. They had concerns for instability, delayed injury, and repeated injury. When the neurologic deficits have resolved, the authors suggest to consider a hard collar for 1-2 weeks with regular clinical reevaluation rather than prolonged immobilization. (16) In a multi-center study by Mahajan et al. no patient with normal MRI (Real SCIWORA) underwent surgery whereas 6-13% of the other "SCIWORA" patients received halo immobilization or internal stabilization. 20% of the Real SCIWORA patients did not receive bracing after discharge.¹³

Pang et al. first description of SCIWORA predated routine MRI and it's ability to identify soft tissue lesions which are not evident on radiographs and CT scans.¹ Evolvement of MRI led to a new understanding of the spectrum of injuries that were subsumed under the name of SCIWORA and the definition of SCIWORA will likely continue to evolve in line with further advances in MRI techniques.¹⁴

20 years after the first publication of SCIWORA, Pang critically re-evaluated the concept of occult instability in the spine of a patient after SCIWORA and described 5 different neural MRT patterns and their clinical outcome: No findings in MRI, spinal cord transection, major and minor hemorrhage and edema. The clinical correlation revealed, that transection and hemorrhage had worse outcome than edema only and patients with no findings in MRI showed most improvement.¹⁶ Therefore identification of "Real SCIWORA" patients—the absence of pathologies even in high-resolution MRI—is valuable because it is highly predictive for an excellent outcome.

The Advanced Trauma Life Support (ATLS) protocol is applied for traumatic spinal cord injuries (SCI), and therefore provides the basis of the treatment algorithm for patients with Real SCIWORA.²⁰

Our standardized protocol involves an interdisciplinary approach: The spine is immobilized, if it wasn't already done in the field. The presence of a trauma surgeon, pediatrician and neurologist is advisable to detect subtle neurologic deficits that may be overlooked, especially in preverbal children. We provided neurologic clinical examination by a specialist in all patients with neurologic deficits and pediatric assistance for all patients under the age of 18. After clinical examination and stabilization of the patient radiologic evaluation was executed. The radiographic algorithm was based on trauma mechanism, age, clinical presentation, and hemodynamic stability. In patients with manifest neurologic symptoms and radiographs without pathologic findings, a MRI scan of the whole spine is mandatory.²¹ The duration of spinal immobilization is dependent on the recovery of neurological symptoms and general clinical presentation but was generally recommended for 2-6 weeks in our population. In literature, there is no evidence for any advantage for longer immobilization in "Real SCIWORA."22,14 Regardless of neurological findings, there is no recommendation for any surgical intervention in "Real-SCIWORA" patients, since any radiologic evidence of pathology is an indication for a more severe diagnosis than "Real-SCIWORA Syndrome."20

Pang et al., as well as several other authors, stated that initial neurological status is the only telltale sign of outcome.^{16,23,24} We reevaluated all patients 6 weeks after trauma and 25/32 after 6.9 years (1-14 years). None of the patients reported any relevant physical residue. In our cohort of only "Real-SCIWORA" patients, we found that not the initial clinical impairment determines the outcome but the absence of pathologies in MRI. If the MRI shows any signs of injury there is a more accurate diagnosis than SCIWORA. In our cohort, a complete recovery was achieved after 3.9 days (1-13 days). This underlines the necessity of a clear diagnosing algorithm including a high-resolution MRI to confirm the diagnosis of "Real SCIWORA" per exclusionem of any other pathology.

To further characterize our patients we also collected data referring to the psychological impact of the trauma with neurological symptoms depression, anxiety and PTSD is well described for SCI.^{25,26,27} The rate of PTSD for SCI is 7-44%.²⁸ In our cohort 4 out of 25 patients reported symptoms of a PTSD even though there was no structural injury and all patients achieved complete recovery of neurologic symptoms. Therefore, we expanded our treatment algorithm and offer a psychologic co-treatment to "Real SCIWORA" patients. Surely, our study has some limitations. The main issue is the limited number of cases and the retrospective design. Even in a large level I trauma center, it was not possible to enroll more than 32 patients to the study. However, the strict inclusion criteria for the "Real SCIWORA" and exclusion of all peripheral reasons for neurologic impairments lead to this small number of cases. However, this is the first reasonable number of cases on mid-term outcome of "Real SCIWORA" in literature. Further studies are needed to substantiate the hypothesis of complete recovery for all cases of "Real SCIWORA" syndrome.

Conclusion

In this retrospective study we report on the largest series of only "Real SCIWORA" patients. With exclusion of any posttraumatic pathology in a high-resolution MRI, we observed full recovery in all patients. Therefore, we advocate for a diagnosis per exclusionem, a full spinal MRI is mandatory. Even though all patients recovered from neurologic symptoms, it is of utmost importance to evaluate each case to ensure detection of structural and potentially serious reasons for the neurologic condition. Psychological impact of neurologic disability and trauma are substantial and psychological support is advisable to prevent PTSD.

Declaration of Conflicting Interests

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Implications and Contributions

VF carried out data interpretation, performed literature research and drafted the manuscript. VA, KB, CS, JK and SL were involved in the design and correction work. FB was significantly involved in design, coordination and draft of the manuscript. All authors red and approved the final version of the manuscript.

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