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Review Article

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Clinical diagnosis and treatment of spinal cord injury without evidence of abnormality in children: a review

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ABSTRACT

Spinal cord injury without radiological abnormality (SCIWORA) is a challenging circumstance in all age groups. Symptoms in some children of SCIWORA does not develop at the time of trauma. Different treatment methods (steroid therapy, immobilization, surgery) applied to treat SCIWORA, but with different prognosis. SCIWORA diagnosed when X-rays and computed tomography (CT) normal; however, initial magnetic resonance imaging (MRI) sometimes can be expected. Performing diffusion-weighted imaging (DWI) with early conventional MRI is a gold standard in the diagnosis of SCIWORA. Early MRI and DWI provides a quick diagnosis to facilitate recovery, ensure a superior clinical outcome, and choose a proper treatment method. Although it provides an accurate diagnosis, early immobilization with steroid therapy can present a good prognosis but not in all cases. Durotomy with duraplasty had a good outcome in adult patients, which might be helpful in children. This review explores the current and future methods of diagnosis and treatment of SCIWORA in children.

Keywords: SCIWORA, MRI, Laminectomy, Durotomy, Duraplasty

INTRODUCTION

Spinal cord injuries (SCI) are scarce in pediatric patients and represent less than 10% of total spinal cord injuries. Therefore, younger patients with SCI are more likely to develop SCIWORA due to the elasticity of the vertebral ligaments and immaturity of bony structures in the pediatric spine. ¹⁻³

Many articles on SCIWORA include when the neurological deficit appeared, when a magnetic resonance imaging (MRI) was performed, and the methods of treatment. However, performing an MRI on admission or within the first seven hours of injury can help in early diagnosis. ^{4–8} T1 and T2-weighted MRIs of the spinal cord differentiate white from grey matter and identify macroscopic anatomical changes within the cord in a diseased or injured area. In pediatrics, various treatment

methods have poor effects on SCIWORA; some authors advocate treating patients with immobilization and corticosteroid therapy followed by rehabilitation. 9–14 The treatment of SCIWORA in children depends on supportive treatment and the prevention of secondary injuries, such as external immobilization to fulfill the patient's mobilization and rehabilitation. This review aims to explore the importance of MRI in the diagnosis of SCIWORA and investigates appropriate treatment methods that will produce a better clinical outcome and quick recovery.

METHODS

In this work, the management and diagnostic methods of SCIWORA in children were reviewed along with appropriate treatments (Figure 2). PubMed, google scholar, academia web, and other websites were used to

select articles relevant to spinal cord injuries without fractures and dislocation in patients between infancy and <18 years old. Excluded articles included those with fractures and dislocation and those referring to patients over the age of 18. Initial research found SCI-related articles (n=140); the inclusion criteria were then applied to find articles correlated to SCIWORA in children (n=24) (Figure 1).

Inclusion criteria included infancy to less than 18 years old, trauma patients with spinal cord injury, no fractures or dislocations on the X-ray or CT scan and articles written in the English language.

Exclusion criteria excluded articles referring to adult patients, X-ray or CT showed abnormality and articles not written in the English language.

Patients data were analyzed using SPSS statistical software (version 22). Treatment methods were compared

along with the improvement in neurological symptoms. The p value was <0.05.

RESULTS

A detailed survey was performed and 24 articles were analyzed that included children with SCIWORA. There

were 177 SCIWORA patients aged from infancy to <18 years old who fulfilled the inclusion criteria (Figure 1). While 119 patients showed an improvement in their symptoms, 58 did not change (Figure 3b).

Patient characteristics included the youngest patient was 12 months old and the oldest was <18; the gender split was 62.5% males and 37.5% females. The most common cause of trauma was an MVA (n=59, 33.33%) (Figure 3d), followed by a fall (n=47, 26.55%), sport (n=39, 22.03%), and other mechanisms (n=32, 18.09%) (Table 1). The most injured regions were cervical, thoracic, and in a few cases, the lumbar region.

Imaging characteristics included the earliest MRI scan was performed on admission, while the latest was carried out on the fourteenth day from the onset of the injury. MRIs play a crucial role in revealing oedema, haemorrhage, and cystic cavities after the injury. Sixty cases showed a normal MRI without any abnormalities or pathological changes. However, using a conventional MRI with other techniques, like DWI, can show the appearance of abnormal signals or detect early signs of ischemia.⁵ The other cases showed oedema, contusion, hyperintensity signals, swelling, or haemorrhage after performing an initial MRI, as presented in Table 2.

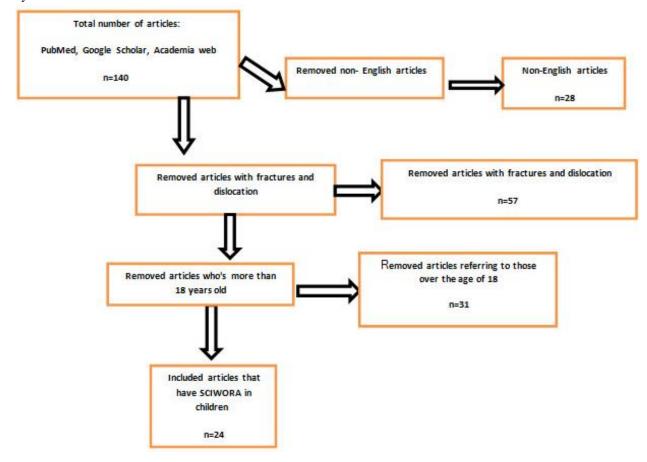


Figure 1: Inclusion criteria for article selection.

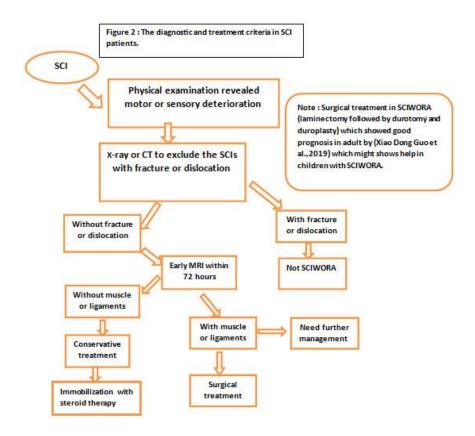


Figure 2: Diagnostic and treatment criteria in SCI patients. After exclusion of fractures and dislocation by X-rays and CTs, early MRI to detect the pathological changes and choose appropriate treatment method.

Table 1: Cases data.

	N, Avg Sex			Trauma mechanism						
Author	age (year)	M	F	Fall	MVA	Sport	Other	Treatment	Prognosis	
Bansal et al (2016) ¹⁵	1 case, 9	0	1	1	0	0	0	Immobilized in pediatric cervical collar, corticosteroids	Good prognosis	
Izma et al (2003) ¹⁰	3 cases, 4	2	1	0	3	0	0	Immobilization	2 cases showed good prognosis; 1 case had a bad outcome	
Beck et al (2000) ²	1 case, 16	0	1	0	0	1	0	High dose methylprednisolone 30 mg/kg	Complete recovery	
Albuja et al (2017) ⁵	2 cases, 14.5	0	2	0	0	0	2	NS	1 case became paraplegic; 2 nd case showed limited improvement	
Feldman et al (2008) ¹⁶	5 cases, 6.1m	0	5	0	0	0	5	Immobilization and hospital course	1 case became normal; others had poor prognosis	
Liao et al (2005) ⁶	9 cases, 4	6	3	4	5	0	0	Immobilization, steroids in 7 patients	7 patients showed good prognosis; 2 remained the same	
Kim et al (2016) ¹⁷	2 cases, 13.5	2	0	0	1	1	0	Immobilization, methylprednisolone	Paraplegic, complete recovery	
Klakeel et al (2015) ¹²	2 cases, 13	2	0	0	0	0	2	Corticosteroids	1 case had a poor outcome; the other no improvement	

Continued.

Author	N, Avg	Sex		Trauma mech		nanism		Tucctment	Duognosia	
Author	age (year)	M	F	Fall	MVA	Sport	Other	Treatment	Prognosis	
Sidram et al (2009) ¹⁸	9 cases, <12	7	2	6	2	0	1	External immobilization, methylprednisolone	7 patents complete recovery; 2 showed partial recovery	
Panagopoulos (2018) ⁷	1 case, 6	0	1	1	0	0	0	Guilford-type brace	Full recovery	
Kriss et al (1996) ¹	3 cases, 2.5m	2	1	0	2	0	1	Conservative treatment	2 patients did not recover; 1 had a good outcome	
Hon et al (2006) ⁴	1 case, 9	0	1	1	0	0	0	Corticosteroids	No recovery	
Mahajan et al (2013) ¹⁹	69 cases, <16	53	16	18	8	27	16	Immobilization and corticosteroids	Most of the patients showed good recovery	
Kim et al (2008) ¹¹	1 case, 1	0	1	1	0	0	0	Neck collar	After follow-up, showed good recovery, near normal	
Kalra et al (2006) ²⁰	1 case, 2.5	1	0	1	0	0	0	Proper physiotherapy was advised	Poor outcome	
Koestner et al (2001) ¹³	1 case, 17m	0	1	0	0	0	1	Cervical collar	Poor outcome	
Pang et al (1989) ²¹	55 cases, 6m-16	29	22	13	31	10	1	Patients initially underwent immobilization	33 children with mild or moderate injuries showed sufficient improvement; 22 children with poor outcome	
Ergun et al (2003) ²²	1 case, 12	0	1	0	0	0	1	Steroids for 6 weeks followed by oral prednisolone	Good recovery	
Silman et al (2008) ²³	1 case, 11m	1	0	0	1	0	0	Conservative treatments	Patient remained paraplegic.	
Matsumura et al (1990) ¹⁴	1 case, 3	0	1	0	1	0	0	Halo vest for 8 weeks	No recovery	
Ayaz et al (2013) ²⁴	1 case, 3.5	1	0	0	1	0	0	Therapeutic exercises and electrical muscle stimulation	Poor recovery	
Sesmat et al (2018) ²⁵	3 cases, 2.3	NS	NS	0	3	0	0	NS	No recovery	
Liang et al (2019) ²⁶	3 cases, 6.33	1	2	1	0	0	2	Corticosteroids, operation	Poor prognosis	
Nagasawa et al (2017) ⁸	1 case, 13	0	1	0	1	0	0	Bed rest, neck collar	Complete recovery	

F=female; M=male; MVA=motor vehicle accidents; NS=Not stated; n=number of SCIWORA cases.

Relationship between the treatment method and prognosis

Fourteen articles reported using immobilization methods to treat SCIWORA, while only 11 articles highlighted the use of corticosteroids; one article focused on the use of corticosteroids with an operation, but this demonstrated a poor prognosis. Combining both immobilization and corticosteroids into one integrated method to treat the patients showed a better clinical outcome than using only immobilization or corticosteroids. ^{6,12,15,17-19}

Two patients received physiotherapy and three patients had an operation and were given steroids, but they did not show any improvements. Five patients received steroid therapy and two of them demonstrated improved symptoms. Sixty-four patients had immobilization only and 39/64 showed an improvement in symptoms. The use of steroids with immobilization was the best treatment method as this showed improvements in 74/98 patients (Figure 3c). Four patients were non-stated (Figure 3a).

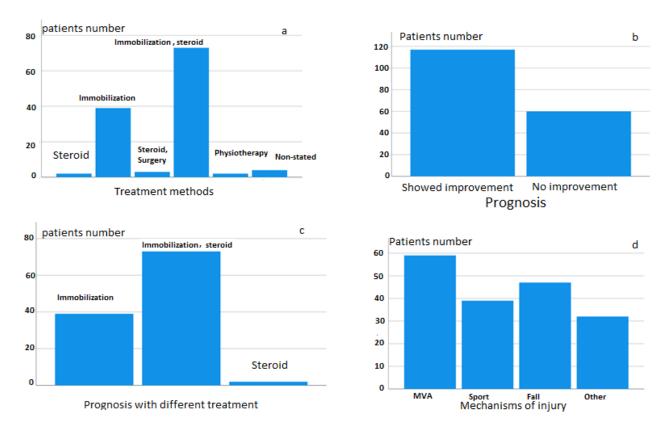


Figure 3: Statistical analysis of all patients included in this review, (a) patient numbers according to the treatment method, (b) patient numbers with improvement in neurological symptoms, (c) neurological improvement after treatment, (d) cause of injury.

Table 2: Images and appearance on MRI.

Study	Level	Time of MRI (days)	Appearance on MRI	X-ray	CT
Bansal et al (2016) ¹⁵	Cervical	NS	Non-hemorrhagic cord edema	NS	Yes
Izma et al (2003) ¹⁰	Thoracic- cervical	6-9 days	Epidural hematoma Atrophic spinal cord Unilateral swelling	Yes	Yes
Beck et al. (2000) ²	Cervical	Next morning after admission	Minimal edema and swelling	Yes	NS
Albuja et al (2017) ⁵	Thoracic Thoracic- lumber	Admission	Hyperintensity	Yes	NS
Feldman et al (2008) ¹⁶	Cervical	2-5	Right posterior medulla hemorrhage, cord swelling	Yes	Yes
Liao et al (2005) ⁶	Thoracic- cervical	3 hours-14	2 spinal cord transection cases 1 edema case 3 contusion with hematomyelia cases, 3 normal cases	Yes	Yes
Kim et al (2016) ¹⁷	Thoracic, cervical	NS	Contusion or hematoma Focal central disc herniation	Yes	Yes
Klakeel et al (2015) ¹²	Thoracic	NS	Hyperintensity ischemic or demyelinating process	NS	Yes
Sidram et al (2009) ¹⁸	6 cervical 3 thoracic	NS	3 normal cases 1 contusion case 5 edema cases	Yes	Yes
Panagopoulos (2018) ⁷	Cervical	Admission	Abnormal signal intensity	Yes	Yes
Kriss et al (1996) ¹	Cervical	NS	Abnormal signal	Yes	Yes
Hon et al (2006) ⁴	Thoracic	Admission	Hyperintensity with contusion	NS	Yes

Continued.

Study	Level	Time of MRI (days)	Appearance on MRI	X-ray	СТ
Mahajan et al (2013) ¹⁹	Cervical	NS	54 normal cases 15 abnormal signal cases	Yes	Yes
Kim et al (2008) ¹¹	Cervical	7 days	High-intensity signal	Yes	Yes
Kalra et al (2006) ²⁰	Cervical	NS	High-intensity signal	Yes	Yes
Koestner et al (2001) ¹³	Cervical	14 hours	Cord edema	Yes	Yes
Pang et al (1989) ²¹	43 cervical 12 thoracic	NS	NS	Yes	Yes
Ergun et al (2003) ²²	Cervical	2	Hyperintensity lesion	Yes	NS
Silman et al (2008) ²³	Thoracic- cervical	NS	Contusion and hemorrhage	NS	Yes
Matsumura et al (1990) ¹⁴	Thoracic- cervical	NS	Abnormal intensity	Yes	NS
Ayaz et al (2013) ²⁴	Thoracic	Next morning after admission	Unremarkable	Yes	Yes
Liang et al (2019) ²⁶	Thoracic- lumber	3 days after injury	Contusion and laceration, T2 hyperintensity	NS	Yes
Nagasawa et al (2017) ⁸	Cervical	Admission	No significant spinal cord lesion	NS	Yes
Sesmat et al (2018) ²⁵	2 thoracic- cervical 1 cervical	NS	2 contusion cases 1 high-intensity signal case	Yes	Yes

CT=Computed tomography; MRI=Magnetic resonance image; NS=Not stated.

DISCUSSION

Pang and Wilberger discovered SCIWORA in 1982 and prescribed it as a spinal cord injury with no evidence of fracture or instability on an X-ray or CT scan.²⁸ It is called real SCIWORA if the evidence is undetectable on an MRI.³⁰ Pang and Wilberger described the mechanisms of neural damage in SCIWORA and suggested that flexion force affects the paediatric spinal cord more than extension force.²⁸ Some cases of SCIWORA caused by flexion-extension forces can cause temporary occlusion of the vertebral arteries and end with infarction.^{22,29}

According to the collected articles, the cervical region was the most commonly affected area because of muscle weakness and inadequate ossification of the structural components of spinal segments, which increase spine flexibility.²³ Another reason was that the size of the head is greater than the rest of the body.²⁵ SCIWORA in the upper neck region is more prevalent in young children who are less than eight years old because the neck is more vulnerable to flexion force, while it becomes more resistant to the possibility of similar injuries after the age of nine.²⁴

Pang suggested that the unique features of the paediatric spine increase the risk of SCIWORA. In children who are <ten years old, the uncinate processes are absent, in addition to the end plate's hypervascular growth zone.²⁷ The unique elasticity of children's spines, such as increased flexibility, muscle weakness, and incomplete ossification of bony components, immensely increase the risk of SCIWORA in children.²⁶ Paediatric SCIWORA requires exceptional management because of its dynamic

nature, its development during the child's life, and anatomical discrepancies.

The prediction accuracy in this review covers the significance of delayed-onset follow-up after SCIWORA. This should be considered thoroughly, primarily due to the variations in each case. This review revealed that cases can develop clinical symptoms within minutes of injury or up to a few days later. This is because of rising intraspinal pressure that happens around 30 minutes after the injury and remains elevated up to seven days, while oedema increases in the first three days after the injury and continues over the next several days. 9,39 It is vitally important to conduct an MRI and DWI as soon as possible to determine the pathological changes in the spinal cord. It should also be noted that acute deterioration is the most common type of neurological deterioration among paediatric patients with SCIWORA, and it appears temporarily within the first hours of injury. A DWI has some advantages in determining the ischemic changes in the spinal cord.

The literature reported 60 cases of SCIWORA in children without signal abnormalities on an MRI (i.e., real SCIWORA). Even with advanced MRI technology, the healthy appearance of neuroimaging remains a clinical episode in children with SCIWORA.²⁰ Nevertheless, MRI has improved the ability to diagnose patients with SCIWORA by enabling the visualization of pathological spinal changes.

Diffusion-weighted MRI is more sensitive if the cause of SCIWORA is an arteriovenous malformation (AVM).^{5,18} However, clinical presentations of spinal cord AVMs can include progressive myelopathy or acute haemorrhage,

such as fistulous AVMs, which are more likely to develop progressive myelopathy that can only be detected by MRI. MRI is very appropriate for the diagnosis of nidus in AVM associated with an increased risk of haemorrhage as it estimates the compressed cord and approves the intramedullary AVMs.³¹ Mulcahey et al examined the accuracy of DWI and concluded that it is more sensitive in detecting cord abnormality, which helps in early and accurate diagnosis, aids in deciding on appropriate treatment, and leads to a positive long-term prognosis.³³

After a few weeks, some cases showed a clinical condition that appeared as ascending myelopathy, ≥four vertebral levels above the initial injury.³² However, some cases showed severe atrophy of the spine after one month of injury. An MRI detected spine abnormalities in some cases, such as cord oedema, contusion, hyperintensity signals, swelling, or haemorrhage. This illustrates the pathophysiology and main features of SCIWORA. The patients who underwent early and appropriate management showed an improvement in their neurological symptoms, which enhanced the probability of neurological recovery and reduced complications.^{2,5,7,8,13,16,24}

MVAs are the most common causes of injury, followed by falls, sports, and other mechanisms (abuse, minor trauma, sudden onset). An excess of sports injuries occurred in adolescents, whereas MVA-related injuries were more common in toddlers. Hence, immobilization was essential to approximate neutral spine alignment in paediatric patients. Most of the cases treated with immobilization and steroid therapy showed a complete improvement as complete recovery can happen after mild or moderate lesions. Some cases showed no symptom improvements, and they remained paralyzed. Treatment outcomes in children with SCIWORA depend on the severity of the injury. For instance, a complete injury with severe lesions will not recover.

The variation of clinical outcomes in SCIWORA depends on the severity of the injury, while the importance of the initial neurological status is considered as a leading prognostic factor for a good outcome.²¹ The best recovery scenarios are expected with patients who have oedema or a cord without contusion or haemorrhage; unfortunately, the worst could be a haemorrhage around the cord.

In adults, many studies suggest the use of posterior laminectomy followed by durotomy with duraplasty, which is an effective method of decreasing intraspinal pressure and improving blood circulation at the swollen cord. ³⁴⁻³⁷ Decreasing the pressure at the injury site helps to increase the blood flow, avoid further injury, which can relive cord oedema, and prevent the progress of spinal cord compartment syndrome. Therefore, treating the compartment syndrome depends on opening all the structures to reduce the pressure at the injury site. Muscle

or ligament disruption can only appear on an MRI, which also increases the pressure at the injury site. Furthermore, increasing motor evoked potential at or below the injury site can predict the increase in spinal cord perfusion pressure (SCPP) and play a role in the assessment of injury severity.³

SCIWORA is diagnosed when X-rays and CTs are normal, while MRIs check the severity of the injury as to whether there are ligament and muscle disruption or not.² If there is no tissue disruption, immobilization and corticosteroids can be a good treatment option as this method improved the symptoms in 23/26 cases.^{6.15–18} Laminectomy followed by durotomy with duraplasty is recommended as a choice of treatment when a ligament or muscle is injured as this can decompress the dura against the swollen cord. The curative effect is still hard to judge after surgery in children, but it improves the prognosis in adults.

Twenty-four patients showed no improvement after immobilization with steroid therapy. To treat severe injury, surgical intervention can reduce intraspinal pressure. Dura is the major cause of spine compression in severe injury, so dural decompression and duraplasty can reduce intraspinal pressure and allow the perfusion pressure to be controlled with <60 mmgH to avoid secondary injury.⁴⁰ Zhu et al performed early durotomy with duraplasty in adult patients and showed significant improvements.⁵⁴ Therefore, hypothesize that this procedure can achieve improvement in children who are severely injured. When starting treatment, the supine position, either operatively or conservatively, can increase the pressure at the injury site and potentially cause secondary injury; the lateral position will avoid increasing pressure.38

Finally, this review has a few limitations, such as the lack of a high standard study that provides sufficient information about MRI findings. Additionally, there was a lack of reports on SCIWORA and the exact method to diagnose or treat patients with positive long-term outcomes. Unfortunately, a lack of such critical details, especially regarding the injury severity of SCIWORA (acute, subacute, or chronic), generated another limitation.

CONCLUSION

Early and appropriate management would increase the efficiency of the healthcare system and bring efforts to fruition. A conventional MRI does not exclude SCIWORA, but using another technique, such as DWI can include it. After confirming injury severity, acute and subacute injuries can be treated conservatively while severe injury needs careful management for symptoms to improve. Surgical intervention may be required if the patient shows ligamentous instability or deformity.

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