Spasticity management in children and adolescents after spinal cord injuries

Evangelia Basina[1], Ioannis S Benetos[2] MD, Dimitrios S. Evangelopoulos[3], Spyridon G. Pneumaticos[4]
[1]*Postgraduate student, Medical School of Athens*
[2]*MD, Academic fellow of Orthopaedic Surgery, 2132086209, ioannisbenetos@yahoo.gr*
[3]*MD, Academic fellow of Orthopaedic Surgery, 2132086209 ds.evangelopoulos@gmail.com*
[4]*Professor of Orthopaedic Surgery, 2132086209 spirospneumaticos@gmail.com*

ABSTRACT

Introduction: Worldwide, the leading causes of death in childhood are injuries and violence. Spinal cord injury (SCI) is a potentially crippling injury which usually results in severe and permanent disability; however, it is relatively rare before the age of 15 years and accounts for only a low proportion of all childhood injuries. SCI and resulting spasticity may cause important loss of functionality. Despite its prevalence, spasticity as a syndrome in SCI patients is not always managed effectively. The aim of this study was to review the management of spasticity in children and adolescents with traumatic SCI. For this reason, a review of the current literature was performed following the PRISMA guidelines and using the online GOOGLE SCHOLAR database and the following keywords: spinal cord injury, pediatric population, spasticity, management of spasticity. Thirty-three studies were finally included in this review. **Results:** TENS (Transcutaneous Electrical Nerve Stimulation), FES (Functional Electrical Stimulation), muscle activation pattern during movement attempts, spinal manipulative therapy, non invasive brain stimulation, aquatic therapy or hydrotherapy, acupuncture, spinal cord stimulation and intrathecal baclofen therapy, Botulinum toxin A and selective dorsal rhizotomy appear to have a positive effect in reducing spasticity. However, the use of cannabinoids does not appear to have a specific effect on the pediatric population. Transplantation of bone marrow nucleated cells (BMNC) and multiple mesenchymal stem cells (MSC) appear to have an important role in treating SCI patients, however, more clinical trials are required.

Key Words: spinal cord injury, pediatric population, spasticity, management of spasticity

Introduction

Worldwide, the leading causes of death in childhood are injuries and violence [1]. Spinal cord injury (SCI) is a potentially crippling injury which usually results in severe and permanent disability; however, it is relatively rare before the age of 15 years and accounts for a low proportion of all childhood injuries [2]. The most common level of SCI in children under the age of 14 years is the upper cervical region, while in older children is the thoracolumbar region [3]. SCI and resulting spasticity may cause important loss of functionality [4].

Spasticity is a feature of upper motor neuron syn-

CORRESPONDING AUTHOR, GUARANTOR
Evaggelia Basina, Postgraduate Student, NKUA
Email: ds.evangelopoulos@gmail.com

drome and is a common but not an inevitable sequelae of spinal cord injury (SCI) [5]. The operational definition of spasticity used in this study includes the following items, on which there seems to be consensus in the literature: (1) increased muscle tone (the tonic stretch reflex): (2) increased tendon reflexes (the phasic stretch reflex): (3) increased exteroceptive reflexes (the flexor reflex): (4) pathologic radiation of reflexes (between spinal segments and over time). In general, spasticity is classified as a symptom of the upper motor neuron syndrome, characterized by an exaggeration of the stretch reflex secondary to hyperexcitability of spinal reflexes. Upper motor neurons originate in the brain and brain stem and project to lower motor neurons within the brain stem and spinal cord. The lower motor neurons are of two types, both of which originate in the ventral horn of the spinal cord: (1) alpha motor neurons project to extrafusal skeletal fibers and (2) gamma motor neurons project to intrafusal muscle fibers within the muscle spindle. In terms of pathophysiology, spasticity in SCI results when a lesion of the CNS interrupts the signals sent via the upper motor neurons to the lower motor neurons or related interneurons [4].

Immediately following a SCI, a period described as "spinal shock" exists whereby the patient presents with flaccid muscle paralysis and loss of tendon reflexes below the level of the lesion. Spinal shock has been reported to last from 1 to 3 days to a few weeks post-injury, after which there is gradual development of exaggerated tendon reflexes, increased muscle tone, and involuntary muscle spasms: the symptoms of spasticity. Symptoms of spasticity experienced by SCI patients following the period of spinal shock negatively affect quality of life through restricting activities of daily living [4].

Despite its prevalence, spasticity as a syndrome in SCI patients is not always managed effectively. This is likely due to the fact that the syndrome can have various presentations. It is recommended to take a step-by-step approach with a hierarchy of treatments [4]. The most conservative tactics are utilized first, with a progression from physical rehabilitation modalities, pharmacologic interventions, injection techniques, intrathecal baclofen, and lastly, surgery [4].

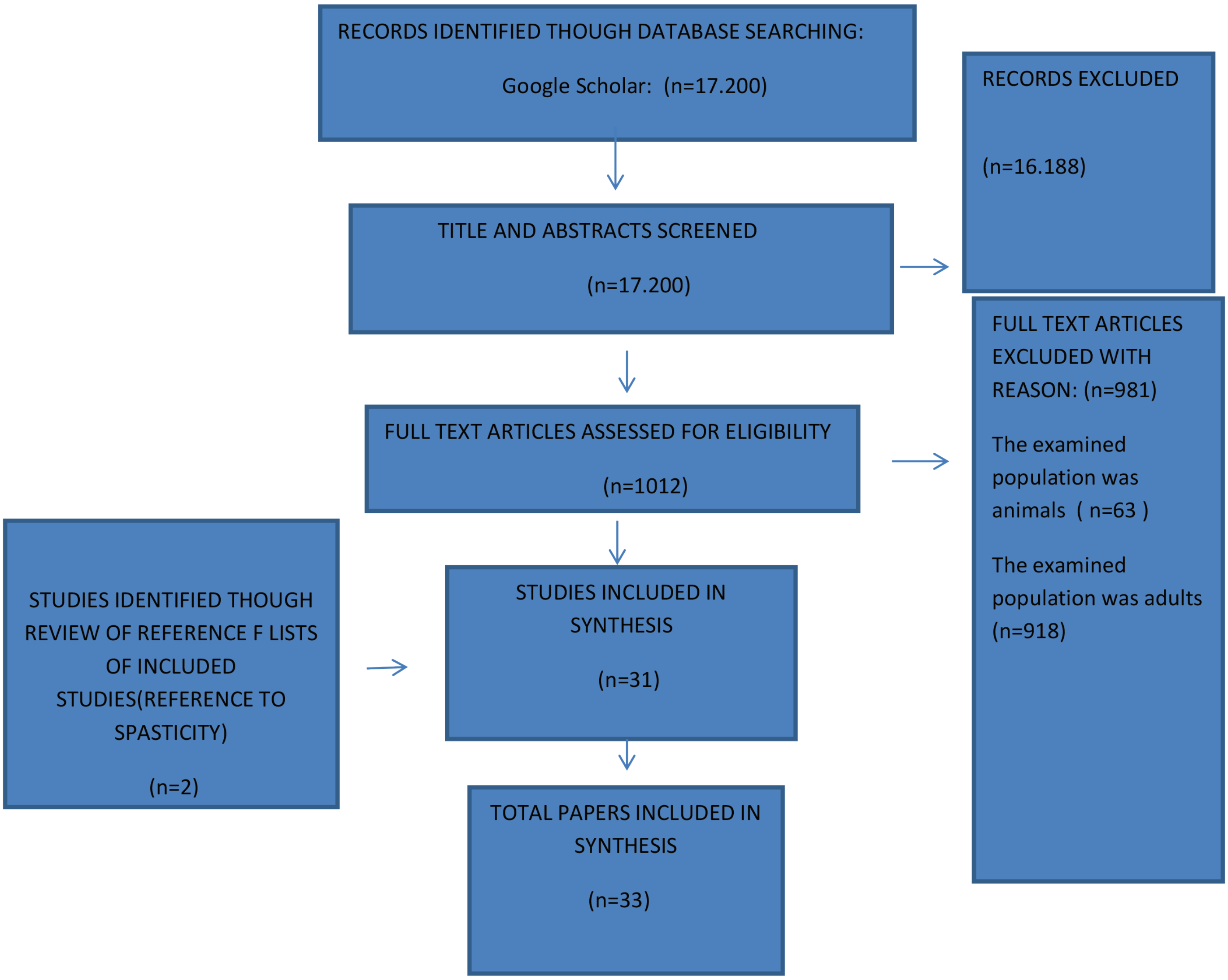
The incidence of pediatric traumatic SCI (0–17 years) in the United States has been estimated to be 17.5 per million-population. The median age at injury is 15 years and the most of patients are boys. No comparative studies have examined whether children with traumatic SCI have a better or worse prognosis than adults with SCI. There were no differences in survival in those injured at ages 0–4 versus 5–9 versus 10–15 years. Life expectancies for those injured before the age of 16 years are reduced compared to both the general population as well as persons with SCI injured at 16 or more years of age. The reasons for this increased mortality and reduced life expectancy are not clear but may be related to a longer exposure to SCI complications and secondary conditions, as well as consequences of developing SCI prior to achieving skeletal maturity [6].

Differences between Pediatric and Adult SCI

The child's cervical spine is characterized by ligamentous laxity, incomplete ossification of the vertebrae, anterior wedging of vertebrae, shallow angle of facets, and relatively underdeveloped neck muscles compared to teenagers and adults. As a result, forces on the head and neck can result in greater stretching of the ligaments and spine without resultant fracture but with injury to the spinal cord. Children with SCI experience some unique secondary complications such as scoliosis and head and neck bone mineral density and other complications such as autonomic dysreflexia require modifications in management due to pediatric physiology. Researchers are evaluating various modes of exercise in youth with SCI, attempting to lessen the impact of SCI on bone, muscle and metabolic health and manage resulting spasticity. The rehabilitation of children with SCI typically includes compensatory strategies and exercise, and new methods of exercise including functional electrical stimulation and activity-based locomotor training are being researched for efficacy in restoration of function [6].

The aim of this study was to review the management of spasticity in children and adolescents with traumatic SCI. For this reason, a review of the current literature was performed by following the PRISMA guidelines and using the online GOOGLE SCHOLAR database and the following keywords: spinal cord injury, pediatric population, spasticity, management of spasticity.

TABLE 1: CURRENT REVIEW FLOWCHART



Inclusion criteria in the review were: primary studies in pediatric population with traumatic SCI published in English language. Initial search resulted in 17.200 articles. After checking titles and abstracts, 16.188 articles were rejected for not meeting the inclusion criteria (studies concerning non traumatic SCI) and the remaining 1012 articles were assessed for eligibility. From them, a great number of articles were also excluded (n=981) due to the fact that they were studying SCI on either animals (n=63) or adults (n=918). Finally, 33 studies were included in the present review, with 2 of them being studies identified thought the review of the reference lists of already included studies (Table 1).

Discussion

Physiotherapy

Transcutaneous Electrical Nerve Stimulation (TENS) is one of the most commonly used methods for the management of neuropathic pain in patients with SCI. This technique is used in combination with massage therapy in order to relief pain caused by spasticity. TENS is useful and safe and when added in long term

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rehabilitation programs can decrease pain to a significant level. In a recent study, a total of 60 patients with incomplete SCI and neuropathic pain were subjected to high frequency TENS (80 HZ). There were two sessions per day with each session lasting for 45 minutes. TENS was applied for four days in a week and all patients were followed for eight weeks. Pain intensity was measured by using VAS (Visual analogue scale). During the consecutive sessions of TENS application, pain intensity decreased in a linear fashion and there was a significant decrease in pain at the end of the treatment [7]. Furthermore, electrical stimulation improves muscle mass and strength, passive range of motion, upper extremity function and walking speed. In addition, positioning of the foot and ankle kinematics during walking, sitting posture and static/dynamic sitting balance may also be improved [8].A recent study showed that Functional Electrical Stimulation (FES) cycling may provide some functional improvement in chronic SCI (more than 2 years duration) after following a program of one hour FES cycling session 3 times per week for 16 weeks. The results showed that there were statistically significant improvements in total motor and FIM scores and spasticity level at the 6 months follow up [9].

Another method of physiotherapy to improve spasticity is muscle activation pattern during movement attempts. In a recent study, the activation pattern included training 2 times per week for 12 weeks on a lower body eccentric resistance (eccentric strength isometric and daily step physical activity). The results showed improvement with eccentric and isometric strength training while daily step activity remained unchanged [10]. In addition, locomotor training in children with SCI (15-18 years old) showed some benefit. Locomotor training included body weight supported treadmill or over ground training [11].

In a recent study, spinal manipulative therapy appeared to be safe and effective, offering pain relief as a result of myofascial release of paraspinal muscles (decrease spasticity) after a treatment with 13 physical therapies over a period of 5 weeks [12].

Non-invasive brain stimulation is also a way to promote motor and functional recovery following SCI. Non-invasive brain stimulation apart from repetitive transcranial magnetic stimulation and transcranial direct current stimulation was used in a research study aiming at motor and functional recovery of spasticity especially in upper and lower extremities following SCI. In the case that the residual corticospinal circuits could be stimulated appropriately, the changes might be accompanied by functional recovery or an improvement in spasticity[13].

In addition, a very important part of physical therapy can be the aquatic therapy or hydrotherapy. Hydrotherapy can be offered to patients with SCI in order to improve gait kinematics, cardiorespiratory and thermoregulatory responses and reduce spasticity [14]. Exercising in the water of a pool has been shown to improve mobility and quality of life and lessen spasticity and pain. It is highly effective in promoting overall recovery from SCI. However, more research is required to thoroughly investigate it and develop protocols and safety measures that will increase the variety of patients with access to aquatic therapy [15].

Acupuncture

Acupuncture has been used to resolve functional recovery problems after central nervous system injury. Researchers suggest that acupuncture has therapeutic potential to help improve limb movement function and decrease the severity of spasticity. Moderate quality evidence suggests that electro-acupuncture combined with conventional routine care (pharmacological/rehabilitation) can reduce spasticity and improve motor function and activities of daily living. In a recent study, 67 SCI patients with lower extremity spasticity were randomly assigned to electroacupuncture and control treatment groups. Electroacupuncture patients received 1-2 Hz for 30 min/day, 6 times/week for 2 months, whereas control received conventional pharmacology. After 2 months of treatment, electroacupuncture decreased lower extremity spasticity in SCI patients and was more effective than conventional therapy. Acupuncture acts on spasticity by breaking pain-spasm cycle, regulating activity of spinal motor neurons and regulating neurochemicals [16].

Pharmacological treatment

A few studies have examined the effects of cannabinoids on spasticity in the pediatric population. However, there is currently insufficient evidence to sup-

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port use of cannabinoids in treatment of spasticity in pediatric patients [17]. In a randomized controlled trial, 72 children and adolescents (8–18y), with spasticity due to traumatic injury were radomly assisted to receive THC (Tetrahydrocannabinol)/CBD (Cannabidiol) or placebo (2:1 ratio) for a period of 12weeks. Up to a maximum of 12 sprays/day, dependent on outcome and tolerance. No difference in spasticity between THC/CBD and placebo were found. Furthermore, were improvements in pain, though not to statistical significance. The posthoc analyses found difference in 12–17y [18]. Cannabinoid use should be discouraged outside well conducted clinical trials. As yet there are no long-term data on the effects of cannabinoids on neurodevelopmental outcome. Parents' reports describe potential cognitive benefits, though effects of the condition on cognition make the effect of CBD on the developing brain difficult to assess. The fact that in the majority of children spasticity results from neurodevelopmental abnormalities complicates the assessment of neurological effects of cannabinoids in pediatric population [17].

Medications such as baclofen that act on the central nervous system and botulinum toxin infusion causing muscle denervation, can be used against spasticity [19]. Baclofen is used for the relief of flexor spasms, clonus and concomitant pain [20]. However, intrathecal baclofen therapy (ITB) has been used in the treatment of spasticity in pediatric population with a delay in referral [21]. Often, only after a long time of failed medical therapy, the use of a baclofen pump is considered [22]. In a recent study, ITB had a dramatic long-lasting effect on spasticity (out of 30 patients, with age ranging from 5 to 23 years old, 20 reported effectiveness of ITB and 26 reported an improved quality of life). Despite the limitations of this study, earlier referral for ITB showed better results in treating severe spasticity. In a recent study, ITB showed very good reduction in spasticity and painful deformities caused by this condition in 80 - 97% of cases [23]. Spinal cord stimulation and intrathecal baclofen therapy are used for patients with severe spasticity after SCI [24].

Botulinum neurotoxin (BoNT) is one of the mainstays in the treatment of pediatric spasticity. It is known that BoNT is effective at reducing spasticity and improving range of motion, but it remains to be determined to what degree this translates into improved function, activity, and participation [25]. A recent study including 6 children with spastic diplegia, assessed the results of the combined use of Botulinum toxin A and electrical stimulation (ES) in treating spasticity. The outcomes were not significant at 4 weeks. The authors concluded that the addition of ES does not improve spasticity over any possible effects that BoNT/A therapy has when used alone [26]. Another study assessed the combination of botulinum toxin (BoNT) injections with virtual reality in the pediatric population for management of spasticity. The results showed that the VR intervention was well tolerated, and patients' guardians requested that it be used again in 9 out of 14 cases. VR was helpful in reducing BoNT procedure-related discomfort in the majority of patients. Challenges with VR setup, patient tolerance, and selecting viewing experiences were identified to guide further research and use of VR in a clinical environment [27].

Orthopedic and neurosurgical treatments

When conservative treatment fails to treat spasticity, surgical treatment such as upper extremity tendon transfers and nerve grafting/transfers can be used [28]. In addition, the selective dorsal rhizotomy technique can also be used for the management of spasticity in children and teens. Nowadays, rhizotomy is the most commonly performed operation to treat spasticity in children and is a reasonable option to consider for relieving spinal related spasticity [29,30]. A study with a 20-year follow-up by the Cape Town group, showed very good long-term outcomes in patients after selective dorsal rhizotomy. In a group of 14 patients, all but one had long-term control of their spasticity and good functional improvement that lasted into adulthood [23]. In another study, selective dorsal rhizotomy permitted the treated patient to acquire better ventral posture, abduction, and sitting posture [31]. Recently, the use of stem cells in treating and improving symptoms of neurological diseases has gained increased interest. The role of these cells in tissue repair by secretion of hormones and growth factors, induction of cell division and differentiation in local cells and stem cells in damaged tissue needs to be thor-

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oughly investigated [32]. The transplantation of bone marrow nucleated cells (BMNC) and multiple mesenchymal stem cells (MSC) have a very important role in treating SCI patients. In a recent study, a 15 year old girl with total spinal cord interruption was treated with BMNC and MSC transplantation followed by an intensive neurorehabilitation treatment. Within 2 years of treatment, the ASIA score improved gradually from A to C/D, and it reflected the sensation level change and the patient's ability to control the body trunk. Most importantly, the patient recovered some of the movement activities in lower parts of the body and gained the ability to stand in a standing frame and was able to walk with the support of hip and knee orthoses. The results were impressive; however, more clinical trials are required [33].

Conclusion

Although the incidence of SCI in children is low compared to adults, there are important differences practitioners should be aware of. Age at injury and skeletal maturity are important factors when considering the child's functional goals and risks for long-term complications such as scoliosis and fractures. Quality of life is shaped by the child's physical health, the secondary complications of SCI the child experiences, and its active participation in the family and community. Physiotherapy and medication but also orthopedic and neurosurgical treatments can be especially helpful in improving spasticity and patients' overall quality of life. Each intervention separately and sometimes their combination can offer effective management of spasticity. Further research into innovative interventions will contribute to better spasticity management.

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