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Management and Outcomes of Traumatic Paediatric Spinal Cord Injuries in Low- and Middle-Income Countries: A Scoping Review

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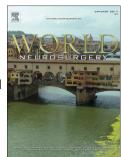
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# Management and Outcomes of Traumatic Paediatric Spinal Cord Injuries in Low- and Middle-**Income Countries: A Scoping Review**

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#### **Abbreviation list:**

TCSI: traumatic spinal cord injury LMIC: low-and-middle income country ASIA: American Spinal Injury Association

#### **ABSTRACT**

# **Background**

Traumatic spinal cord injury (TSCI) is a subset of neurotrauma, which is a significant contributor to global trauma mortality and morbidity in children. The management and outcomes of paediatric TSCI in low-and-middle income countries (LMICs) is unknown. We conducted a scoping review to characterise the methods of management and outcomes of TSCI in LMICs.

#### **Methods**

MEDLINE, EMBASE, and Global Index Medicus were searched from database inception to February 15, 2021. Studies reporting management or outcomes of paediatric TSCI in LMICs were included. Pooled statistics were calculated using measures of central tendency and spread.

#### **Results**

A total of 1171 studies were identified, of which, 5 were included. A total of 212 patients were included in our review with age of participants ranging from 2.5 to 18 years old (mean = 15.4 years). Most patients were male (n=162, 76.4%). The commonest cited cause of injury were falls (n=104/212, 49.1%). The most common level of injury was cervical (n=83, 39.2%). The majority of patients underwent surgery (n=134/212, 63.2%). The extent of injury was quantified and classified using the ASIA chart in only one paper. Long-term management data was not present in any of the included studies.

#### Conclusion

There is a scarcity of published studies reporting the management and outcome of paediatric TSCI in LMICs. The paucity of studies in this domain provides insufficient data to be compared, reducing the ability to draw a strong conclusion. This hinders the development of guidelines to inform best practice.

#### INTRODUCTION

Trauma is responsible for over 5 million deaths annually and accounts for an estimated 20% of total disability-adjusted-life-years (DALYs) lost globally. This is especially apparent in low- and middle-income countries (LMICs), where mortality from trauma is twice that of high-income countries (HICs) and responsible for 15% of the total disability-adjusted-life-years (DALYs) lost globally. Neurotrauma, defined as injury to the brain and/or spinal cord, is a significant contributor to global trauma mortality and morbidity. Long-term effects of neurotrauma are common and extensive, affecting both the patient and their family. These include physical disability, psychological harm, and financial burden. The impact is especially profound when neurotrauma occurs in the paediatric population. The inordinately high number of DALYs lost makes paediatric neurotrauma a global health priority. 3,4

Traumatic spinal cord injuries (TSCIs) are a subset of neurotrauma. Up to 5% of all TSCIs occur in children.<sup>5</sup> According to the 2016 global burden of disease, the incidence and prevalence of paediatric TSCI are 0.3 million and 3.2 million.<sup>3</sup> Common causes include road traffic accidents, falls from height, and sporting injuries.<sup>5</sup> In comparison to the adult population, paediatric TSCIs tend to result from different mechanisms, owing in part to anatomical differences in ligaments and relative head to neck size.<sup>6</sup> In addition, the long-term consequences of injury may differ in comparison to adults; there is greater potential for neurological recovery in children, but higher risk of subsequent scoliosis, particularly if the injury occurs before their adolescent growth spurt.<sup>7-9</sup>

Early and effective management of paediatric TSCI is key to improving patient outcomes in this population. Optimal acute management includes early immobilisation of the spine with potential surgical stabilisation.<sup>6</sup> The extent of injury is often quantified and classified using the American Spinal Injury Association (ASIA) chart.<sup>10</sup> Long-term management is essential and aims for rehabilitation. This is best achieved through a dedicated multi-disciplinary team, including physiotherapists, occupational therapists, and psychologists.<sup>6</sup> Given resource limitations in LMICs, providing effective paediatric TSCI care may be challenging; especially as TSCI is more common in LMICs.<sup>11</sup>

To elucidate the management and outcomes of paediatric patients with TSCI in LMICs, we conducted a scoping review. The primary aim of the scoping review was to characterise the methods of management of TSCI in LMICs. The secondary aim of the scoping review was to determine the outcomes of patients that underwent each management strategy. This scoping review was conducted with the hope of informing the development of future TSCI guidelines pertaining to children in LMICs and moving closer towards the Sustainable Development Goal 3.2 of ending preventable deaths of children by 2030.<sup>12</sup>

#### **METHODS**

A scoping review on the management and outcomes of paediatric traumatic SCIs in LMICs was conducted as per the published protocol.<sup>13</sup> The Arksey and O'Malley scoping review framework was used

to guide the scoping review. <sup>14</sup> The Preferred Reporting Items for Systematic Review and Meta-Analysis extension for Scoping Reviews (PRISMA-ScR) guidelines were used to report the findings. <sup>15</sup>

#### Inclusion and exclusion criteria

We included studies that fulfilled the specific inclusion criteria discussed in our published protocol. <sup>13</sup> For the purposes of this review, TSCI is defined as "external injury to the vertebral column which leads to mechanical compression or distortion of the spinal cord leading to diminished or lost function below the spinal cord level of the injury", paediatric populations as individuals aged between 0 and 18 years <sup>16–18</sup>, and LMICs are those defined by the World Bank Criteria. <sup>19</sup> Prior to the commencement of this study, all reviewers attended an online training and support session to ensure an accurate and standardised approach to the overall methodological process.

There were no restrictions to the type of studies that were eligible for inclusion. Publications in either English or French languages were considered. Studies that (a) did not include paediatric populations (or did not have disaggregated data about a paediatric population), (b) did not discuss TSCIs (or did not have disaggregated data about TSCIs), (c) were neither written in English nor French (this was later redacted), and (d) were not related to LMICs (or did not have disaggregated data about a LMIC population) were excluded.

#### Search strategy

The search protocol for this scoping review was executed in MEDLINE, EMBASE and Global Index Medicus covering the period between database inception to 15<sup>th</sup> February 2021. The search strategy used variants and combinations of search terms related to TSCI, paediatric populations and geographical areas classified as LMICs. Appendix 1 shows the exact content and order of the search string queries.

# **Study selection**

All the articles resulting from the search were exported into Rayyan, where duplicates were identified and deleted.<sup>20</sup> The study selection process consisted of multiple steps. Firstly, two reviewers of SB, DUD, SS, USK, NDAB, AE, JK and GH independently screened the titles and abstracts of the identified articles based on the pre-defined inclusion and exclusion criteria. Any disagreement between the two reviewers' decisions prompted further discussion. If a disagreement persisted, a third reviewer (SB, USK or NDAB) was sought to resolve the disagreement. The full texts of the remaining articles were retrieved and screened by two reviewers independently. Following completion of the study selection process detailed above, all studies initially excluded due to language restrictions underwent full-text screening after being translated using Google Translate.

#### **Data extraction**

Prior to data extraction, an Excel proforma sheet was used to ensure all participants in the data extraction step were extracting data homogeneously. The Excel sheet included columns of specific interest for data extraction such as study design, patients' demographics/characteristics, cause of injury, type of intervention and outcomes of care. Data extraction was performed in two stages, a pilot stage followed by a proper stage. The pilot stage consisted of having multiple authors (DUD, SZYO, SB, AE, NDAB and JK) each going through the same 10 selected articles to extract data. This was to ensure that all participant

authors were able to extract data accurately for a swift data analysis stage. It was also important to pilot this stage to ensure the data collection sheet was reflective of the included studies.

### **Data analysis**

Extracted data was analysed by SPSS v.26 (IBM, USA). Pooled statistics were calculated using measures of central tendency and spread. Further statistical analyses were not possible due to the heterogeneity of data from the different studies and the lack of common data elements. The Cochrane guidelines for reporting null reviews was utilised to report the results of this review.<sup>21</sup>

#### **RESULTS**

The database searches identified 1659 records. 1171 records (70.6%) remained after removing duplicates. 926 records were excluded at the title and abstract screening stage (55.8%), and a further 240 papers (14.5%) were excluded at the full text screening stage. The most common reason for exclusion of the articles were that there was no disaggregate data (68.3%) for the paediatric population, followed by no disaggregate data on spinal cord injuries (21.7%). Five articles remained for data extraction (0.3%) (**Figure 1**). 124 articles excluded based on language restrictions at the title and abstract screening stage underwent full-text screening to identify other relevant articles. All of these articles were excluded: 10 were abstract only, 4 had no original data, 5 had no outcomes of interest, 13 had no disaggregated data on paediatric patients, and 92 papers did not have disaggregated data on traumatic spinal cord injury.

The included articles comprised data on TSCI management in five LMICs: Tanzania, Morocco, Senegal, Pakistan and India (**Table 1**). The articles were published between 2000 and 2020 with a peak between 2010 and 2019 (n=3/5, 60.0%). The majority of the articles were case series or case reports (n=4/5, 80.0%).

212 patients were included in this review. The reported age of participants in the studies ranged from 2.5 to 18 years old (**Table 2**). The overall mean age of the children was 15.4 years old and most were males (n=162/212, 76.4%, 95 CI% = 70.7-82.1%). The cause of paediatric TSCI was reported in all 212 of the included cases. Falls were the most common cause of TSCI (n=104/212, 49.1%, 95 CI% = 42.3-55.8%). Other causes of TSCI were road traffic accidents (n=84/212, 39.6%; 95 CI% = 33.0%-46.2%), sport injuries (n=15/212, 7.1%, 95 CI% = 3.6-10.5%), and others (n=9/212, 4.2%, 95 CI% = 1.5-7.0%) (**Table 3**). Only one case of penetrating injury (0.5%, 95 CI% = -0.4-1.4%) was reported<sup>22</sup>; the remaining 211 cases (99.5%, 95% CI = 98.6%-100.5%) were due to blunt force injuries. There were 83 cervical injuries (39.2%, 95 CI% = 32.6-45.7%), 59 thoracic (27.8%, 95 CI% = 21.8-33.9%), 44 lumbar (20.8%, 95 CI% = 15.3-26.2%), 21 thoracolumbar (9.9%, 95 CI% = 5.9-13.9%), two cervicothoracic (0.9%, 95 CI% = -0.4-2.2%) and one sacral (0.5%, 95 CI% = -0.4-1.4%) case(s) in total. The level of injury was not reported in two cases.

All 5 of the studies reported on the setting where the patient received further management: 2 spinal/orthopaedic rehabilitation units, 1 tertiary care center, 1 teaching hospital and 2 military hospitals. <sup>22–26</sup> Only one of the included articles described the healthcare professionals involved in the patients' care. The reported healthcare professionals were orthopaedic surgeons, neurologists, occupational therapists, physiotherapists, and nurses. <sup>22</sup> One study reported on pre-hospital management: one patient (0.5%) had

their C-spine stabilised by a cervical collar. The mean delay to treatment was reported in three studies; this ranged from 1 hour to 2.5 years.

The hospital management of paediatric TSCI was reported in 210 (99.1%, 95 CI% = 97.8-100.4%) of the included cases. 75 patients (35.4%, 95 CI% = 28.9-41.8%) were treated conservatively – including the patient who suffered a penetrating injury – and 134 patients underwent surgery (63.2%, 95 CI% = 56.7-69.7%). Only three surgeries were described in detail; all operations were posterior approach arthrodesis.

Complications of conservative treatment was reported in two studies; this included a spastic neurogenic bladder, a gluteal pressure sore, and a urinary tract infection (UTI) due to *Escherichia coli*.<sup>22,25</sup> Aggregated data on complications of both surgery and conservative treatment were provided in one other study; pressure sores and UTIs were reported (Table 4).<sup>26</sup> Only one study reported ASIA scores (before/after) - ASIA A (132/127), ASIA B (19/16), ASIA C (14/22), ASIA D (16/13), ASIA E (23/26).<sup>26</sup> Bansal et al. also reported that 126 patients had a poor outcome (dependent), 51 had a fair outcome (partially dependent), and 27 had a good outcome (independent) at discharge.<sup>26</sup> Diop et al. reported four patients who had an improvement in neurological function at a mean 4.25 months (range: 3-5 months) follow-up.<sup>25</sup> Overall, 4 patients (0.02%, 95 CI% = 0-0.04%) died, although the cause of death was not due to TSCI. The causes reported were: multiorgan failure (1 case), head injury (1 case), and nosocomial pulmonary infection (1 case); one case was unspecified. The deaths reported occurred prior to any surgical management.<sup>22–24</sup> Follow-up length was poorly reported in the other four papers.

#### **Discussion**

Our scoping review is the first to summarise the published literature on the management and outcomes of paediatric TSCIs in LMICs. Previous global TSCI reviews have been conducted in the adult population. <sup>27,28</sup> Compared to these reviews, fewer paediatric TSCI publications were identified and included in our study. This is likely to be due to a global scarcity in clinical research on paediatric spinal cord dysfunction. <sup>29</sup> A recent report concluded that only 25 studies had been conducted on the incidence and aetiology of paediatric spinal cord dysfunction globally; 14 of these studies were regarding paediatric TSCI, but they were predominantly conducted in HICs. <sup>29</sup> Therefore, there is little to no data on the incidence and aetiology of paediatric TSCI in LMICs. It should be noted that they used an age cut-off of 15 years, which may explain why our higher age cut off of 18 years found 5 studies that reported the aetiology of paediatric TSCI. <sup>29</sup> Despite having a higher number of cases, given the relatively few studies conducted in this field, and the paucity of high-quality studies and the differing methodologies, it would not be appropriate to go into a detailed discussion about possible explanations for the differences in aetiology between countries. More, better quality studies are required before this would be appropriate.

The dearth of data on paediatric TSCI in LMICs contrasts with the abundance of studies being conducted on paediatric traumatic brain injury (TBI) in LMICs.<sup>30</sup> It is critical that similar research starts to occur not only on the incidence and aetiology of paediatric TSCI, but also its management. Research on the management of paediatric TSCI and the long-term outcomes of management strategy would help us understand the implications of instrumentation on growing children: how it affects their growth potential and prognosis. Furthermore, investigating management techniques would inform us about resources available in LMICs and their ability to perform safe and timely surgery. This is important because any injury to the spinal cord requires immediate and effective treatment, as patients could suffer from

paralysis. A lack of appropriate equipment increases the likelihood of complications, including death. The first step in solving such an issue would be to conduct research to identify unmet resource needs. In addition, research on TSCI needs to take into account the health professionals involved in the management and rehabilitation of the patients. None of the papers included reported the presence of psychologists, despite the evidence base supporting their role in reducing psychological trauma, which can be especially debilitating at an early stage of life.<sup>31</sup> We propose an urgent study is needed that combines the expertise of the researchers and centres involved with the Global Neurotrauma Outcomes Study for adults with traumatic spinal injury<sup>32</sup>, and the Global Children's Non-Communicable Diseases Collaborative.<sup>33</sup> A significant strength of this study is that it is a scoping review. Therefore, its primary purpose is to chart all existing evidence surrounding this topic; our finding of current scarcity of research in this field should incentivise funding in this area of unmet need for a disease with serious repercussions.

In addition to the need for further research in paediatric TSCI, the selection of appropriate reporting measures needs greater attention from the scientific community. Particularly if study findings are to be useful, reliable and relevant to patients, healthcare professionals and others making decisions regarding healthcare provision.<sup>34</sup> A 2014 Lancet Series 'Research: Increasing Value, Reducing Waste' estimated a staggering 85% of biomedical research offering actual or potential clinical benefit was prevented due to research inefficiency, equating to \$200million lost in 2010.<sup>35</sup> One of the reasons cited for this inefficiency was heterogeneous data collection and reporting. This is further demonstrated by the most cited Cochrane Reviews stating problems due to inconsistencies in the outcomes reported in included studies.<sup>36</sup> The scientific community is enacting increasingly rigorous research methodology and responding appropriately to counteract this wastage through the development of Common Data Elements (CDEs) and Core Outcome Sets (COS).<sup>37</sup> Use of CDEs and COS in research enables the standardisation of terms, facilitates the comparison and combination of data across studies, reduces selective outcome reporting, and improves generalisability of research findings.<sup>37</sup> There is a need for CDEs and COS to be developed and mandated in paediatric TSCI research. For instance, measures such as ASIA should be reported in all studies concerning TSCI management and outcomes, given its significant role in prognostic implications and practical clinical utility. 10,38 This would reduce research wastage in a field where data is already lacking. Research wastage would also be reduced by studies providing disaggregated data or publishing their raw data on data repositories.<sup>39</sup> Both heterogeneous data reporting and aggregation of data were issues identified during the course of this review.

Our review does have limitations. Firstly, studies were initially excluded if they were not written in English or French. This criterion was redacted following the finding that only 5 studies – 4 of which were case reports or case series – were eligible for inclusion. Studies written in other language were translated using Google Translate, which could have resulted in subtle nuances being missed. However, it should not have altered our ability to evaluate whether the paper related to paediatric populations and traumatic spinal cord injuries. Therefore, we are confident that papers written in languages other than French or English were appropriately screened. Secondly, our study did not include grey literature. It is possible that outcomes of traumatic spinal cord injuries may be present in reports from charities or governments. Thirdly, our study is akin to an empty review, as it found few studies of relevance. However, the value of empty reviews has long been noted<sup>21,40–42</sup>. The impetus is now on researchers in this field to collect the relevant data to ensure a future review in this topic finds more studies of note. Researchers should be urged to publish their findings in this field even if their results show no significant findings, as it will

provide a field that is currently reliant on expert opinion<sup>43</sup> with a greater evidence base. A final limitation of our study was that we did not conduct a search on Cumulative Index to Nursing and Allied Health Literature (CINAHL). This may explain the lack of reports of the involvement of allied healthcare professionals in the management of TSCIs in our review. However, similar studies should have been indexed in the databases that were searched. All in all, our study fulfils the role of a scoping review, as it charts the evidence base in this field.<sup>14</sup>

#### Conclusion

This scoping review provides an overview of the management and outcomes of TSCI in the paediatric population in LMICs. Our review found that the most common mechanism of injury was falls and the majority of patients suffered injuries at the cervical region. There was a broad mix of healthcare professionals who were involved in the care of TSCI patients, although this was only reported in one study. Most patients received treatment at a hospital; of these, many were surgically treated. Mortality rate was low, however, there were many reports of complications due to treatment; this was observed in both conservative and surgical management. Of note, there was a significant variation in the mean delay to treatment among reports; the upper limit being 2.5 years. This could explain the high complication rates seen in this study as TSCIs often require immediate intervention.

Our review also found that there is a lack of published literature on the management of this area of interest, with no type 1, 2 or 3 evidence reported. The paucity of studies in this domain provides insufficient data to be compared, reducing the ability to draw a strong conclusion. This hinders the development of guidelines to inform best practice. The lack of guidelines may lead to issues such as a variability of management largely based on a clinician's judgement, which may put patients at a risk of receiving suboptimal or outdated treatment, and/or being exploited with overtreatment or exorbitant healthcare fees. There is a need for research on TSCI in LMICs, especially in the paediatric population for better management of this condition. It is evident from this review that the development of COS and CDEs would help. For example, without a well-defined reporting of the types of spinal cord injuries, a consensus on the management cannot easily be attained. High-quality research will open a window for the development of guidelines for better management.

# **Figures and Tables**

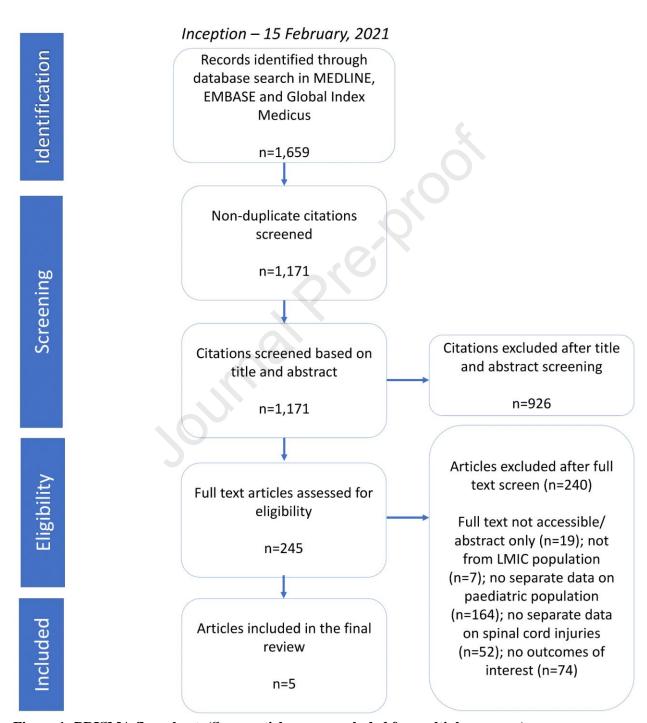


Figure 1: PRISMA flow chart. (Some articles were excluded for multiple reasons.)

Table 1: Characteristics of the 5 included studies

Author, year (study ID)	Country being studied	Study design	Study setting	Number of paediatric TSCI patients
Bansal (2020) <sup>18</sup>	India	Retrospective cohort study	Tertiary Care Centre, Indian Spinal Injury Centre (ISIC), New Delhi	204
Diop (2006) <sup>19</sup>	Senegal	Case series	Department of Neuro- Traumatology, Main Hospital of Dakar	5
Ayaz (2013) <sup>20</sup>	Pakistan	Case report and literature review	Military rehabilitation hospital	1
Nebhani (2015) <sup>21</sup>	Morocco	Case report and literature review	Pediatric Emergency Department of Military Hospital, Rabat, Morocco	1
van Adrichem (2019) <sup>22</sup>	Tanzania	Case report	Kilimanjaro Christian Medical Centre (KCMC), Orthopaedic Rehabilitation Unit, Tanzania	1

Table 2: Characteristics of included participants in the 5 included studies

Author, year (study ID)	Inclusion criteria	Number of paediatric TSCI patients	Age range of paediatric TSCI patients, years (mean)	Number of male paediatric TSCI patients (% of total)
Bansal (2020) <sup>18</sup>	Patients (up to 18 years) with an SCI diagnosis admitted at ISIC between 2002 and 2015	204	3 to 18 (15.7)	157 (76.8)
Diop (2006) <sup>19</sup>	Children with SCI admitted to the Neuro- Traumatology department of the Main	5	2.5 to 14 (8.3)	3 (6.0)

	Hospital of Dakar between January 2004 to October 2005			
Ayaz (2013) <sup>20</sup>	Pre-school child who developed SCIWORA following a road traffic accident	1	3.5 (3.5)	1 (100)
Nebhani (2015) <sup>21</sup>	Patient aged 3 years with an SCI caused by a motor vehicle accident	1	3 (3)	0 (00)
van Adrichem (2019) <sup>22</sup>	Patient aged 17 years with penetrating SCI admitted at KCMC	1	17 (17)	1 (100)
Total		212	<b>(</b> 0)	162 (76.4)

Table 3: Causes of paediatric traumatic spinal cord injury in the 5 included studies

Author, Year (Study	Mechanism of injury, n			
ID)	Road traffic accident	Sport injuries	Falls	Others
Bansal (2020) <sup>18</sup>	79	15	103	7 (not specified)
Diop (2006) <sup>19</sup>	3	0	1	1 (overturned dugout)
Ayaz (2013) <sup>20</sup>	1	0	0	0
Nebhani (2015) <sup>21</sup>	1	0	0	0
van Adrichem (2019) <sup>22</sup>	0	0	0	1 (shot by arrow)
Total	84	15	104	9

Table 4: Complications of injuries in the 5 included studies

Author, Year (Study ID)	Complications of associated injuries, n		
	Pressure sore	Urinary tract infection	Spastic neurogenic bladder
Bansal (2020) <sup>18</sup>	15	5	-

Diop (2006) <sup>19</sup>	1	1	-
Ayaz (2013) <sup>20</sup>	No complications reported		
Nebhani (2015) <sup>21</sup>	No complications reported		
van Adrichem (2019) <sup>22</sup>	1	1	1
Total	17	7	1

#### **Conflicts of interest**

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

#### **Contributorship statement**

**David Ulrich Dalle:** conception, design, screening of data, data extraction, data analysis, writing, reviewing and editing, project administration, visualisation

**Smrithi Sriram:** conception, design, screening of data, data extraction, data curation, data analysis, writing, reviewing and editing, project administration

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**Abdullah Egiz:** screening of data, data extraction, writing, reviewing and editing **Jay Kotecha:** screening of data, data extraction, writing, reviewing and editing **Ulrick Sidney Kanmounye:** conception, screening of data, reviewing and editing,

George Higginbotham: screening of data, writing

**Setthasorn Zhi Yang Ooi:** data extraction, data curation, data analysis, writing, reviewing and editing, visualisation, supervision

**Nourou Dine Adeniran Bankole:** screening of data, data extraction, data analysis, writing, reviewing and editing, validation, supervision

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#### **Ethical approval**

Ethical approval was not required.

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# **Abbreviation list:**

TCSI: traumatic spinal cord injury LMIC: low-and-middle income country ASIA: American Spinal Injury Association

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