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CASE SERIES

The risk of delayed spinal cord injury in pediatric spinal deformity surgery

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Abstract

Delayed spinal cord injury (SCI) hours or days after surgery, with uneventful monitoring and initial normal postoperative neurological examination, is a rare complication. Based on anecdotal evidence, the risk of delayed spinal cord injury might be higher than previously assumed. Therefore the aim of this study was to determine the risk of delayed SCI after pediatric spinal deformity surgery between 2013-2019 in the Netherlands. The total number of pediatric spinal deformity surgeries performed for scoliosis or kyphosis between 2013–2019 was obtained from the Dutch National Registration of Hospital Care. All eleven Dutch hospitals that perform pediatric spinal deformity surgery were contacted for occurrence of delayed SCI. From the identified patients with delayed SCI, the following data were collected: patient characteristics, details about the SCI, the surgical procedure, management and degree of improvement.

2884 pediatric deformity surgeries were identified between 2013–2019. Seven patients (0.24%) with delayed SCI were reported: 3 idiopathic, 2 neuromuscular (including 1 kypho-scoliosis) and 2 syndromic scoliosis. The risk of delayed SCI after pediatric deformity surgery was 1:595 in idiopathic scoliosis, 1:214 in syndromic scoliosis, 1:201 in neuromuscular scoliosis. All seven patients had a documented normal neurological examination in the first postoperative period; neurological deficits were first diagnosed at a median 16h (range 2.5-40) after surgery. The risk of delayed SCI after pediatric deformity surgery is higher than previously reported, especially in patients with non-idiopathic scoliosis. Regular postoperative testing for late neurologic deficit should be performed for timely diagnosis and management of this devastating complication.

Keywords Scoliosis · Spinal deformity · Pediatric · Spinal cord injury · Delayed neurological deficit · Complications

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Introduction

Pediatric spinal deformities are relatively common. With an estimated prevalence of 1–3% idiopathic adolescent scoliosis (AIS) is the most frequently diagnosed form of pediatric deformity [1, 2]. Fortunately, most patients can be treated nonoperative and surgery is only required for severe curve progression [1, 3]. Nevertheless, just for AIS more than 5000 scoliosis corrections are annually performed in the United States alone [4].

Pediatric deformity surgery is challenging and is among the most invasive surgeries performed in children [5, 6]. Complication rates for pediatric spinal deformity surgery have been reported as high as 22% and 7.6% for AIS specifically; respiratory failure, reintubation, and implant-related complications are some of the most commonly reported complications [3, 4, 7]. Neurological deficit due to spinal cord injury (SCI) is, however, perhaps the most feared

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complication of pediatric deformity correction by patients, their family and caregivers [3]. According to the most recent analysis of the Scoliosis Research Society Morbidity and Mortality Database which included 84.320 procedures, the risk for new neurological deficit after AIS surgery is 0.35%. However, reported risks range from 0.2 to 0.7%, the majority of the injuries are incomplete and at least partially recover in the months after surgery [3]. Most common causes are direct trauma to or compression of the cord due to implant malposition (screw or hook, stretch by the correction manoeuvre or ischemia of the spinal cord [6, 8]. The early SCI is usually identified during surgery by the multimodal intraoperative neuromonitoring or immediately after surgery at the postoperative neurological examination [6]. Nevertheless, a much more uncommon form of neurological deficit has also been reported, delayed SCI.

Delayed SCI is defined as a neurological deficit that develops hours to days after uneventful surgery with stable neuromonitoring and a normal postoperative neurologic examination [6, 7, 9–12]. Delayed SCI typically develops within 4 days after surgery and is in 74–86% of the cases incomplete [6, 7]. The reported incidence of delayed SCI ranges from 1:1000 in a single center cohort study to 1:10.000 in a survey among the Scoliosis Research Society members [6, 7]. Based on local anecdotal evidence the suspicion arose, however, that the risk of delayed SCI after pediatric deformity surgery might be higher than previously assumed [6, 7]. Therefore, the aim of this study was to determine the risk of delayed SCI after pediatric spinal deformity surgery in a retrospective national cohort study in the Netherlands.

Methods

Identification of delayed SCI

Pediatric spinal deformity surgery in Netherlands is highly organized and performed in four large non-academic and seven academic hospitals. The spine surgeons of these 11 hospitals were contacted through email and phone to inquire whether they had a case of delayed SCI between 2013 and 2019 in patients < 18 yrs. Delayed SCI was defined as a spinal cord injury that developed at least 1 h after spinal deformity surgery, with uneventful intraoperative neuromonitoring and normal postoperative neurological examination [6, 7]. In the hospitals that administer postoperative epidural analgesia, only patients with normal postoperative neurological examination and abnormal neurological examination after elimination of the analgesia were included. Surgeons that encountered the complication were asked to complete a questionnaire for each identified case. After completion of the manuscript, the abstract was send to all Dutch pediatric spine deformity centers as a final check for missing cases before submission for publication. The study was approved by the medical ethical committee MEC-2022–0427.

Questionnaire

The questionnaire was based on the SRS 1999–2009 questionnaire used by Auerbach et al. [7]. The questionnaire consisted of a case form and a description of the surgical team (Tables 1, 2, 3, 4). The case form included patient characteristics, description of the performed surgery and details about the SCI (Tables 1, 2, 3).

National database

The total number of pediatric spinal deformity surgeries performed for scoliosis or kyphosis between 2013 and 2019 was obtained from the Dutch National Registration of Hospital Care. The Dutch Hospital Data foundation was established to manage, monitor and maintain hospital data and to provide information on hospital care in a National Registration of Hospital Care [13]. This database contains information on all hospital admissions and includes patient demographics, diagnosis based on international classification of disease (ICD) codes and performed operations. Surgical procedures

Table 1 Patient characteristics

	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Median
Age	14 yr	16 yr	17 yr	7 yr	16 yr	17 yr	14 yr	16 yr
Gender	Female	Male	Male	Female	Female	Female	Male	
Type of Deformity	AIS Lenke 3	NM	NM	Syn	AIS Lenke 1A	Syn	AIS, Lenke 3	
Cobb angle	70°	80°	51°*	70°	52°	53°	67°	67°
Co-morbidity	None	Friedreich's ataxia, cardiomyopathy	Double Chromosome 14	Spondylocarpotarsal synostose	DM Type I	Trisomy 21, TSC, Renal failure	None	

AIS adolescent idiopathic scoliosis, *NM* neuromuscular scoliosis, *Syn* syndromic scoliosis, *TSC* tuberous sclerosis complex

*Also 75° kyphosis

Table 2 Surgery details

	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Median
Surgery	Primary case PF T2-L2 3×SPO	Primay case PF T2-L4	Primary case PF T4-T12 6×SPO	Primary case PF T2-L2 6×SPOTraction	Primary case PF T4-T12	Primary case PF T3-L4	Primary case PF T4-L3	
Neuro-monitoring	No change	No change*	No change	No change	No change	No change	No change	
Surgical time	367 min	250 min	270 min	230 min	161 min	319 min	223 min	250 min
Intra-operative blood Loss	600 ml	500 ml	600 ml	640 ml	300 ml	800 ml	550 ml	600 ml
Postoperative blood loss (drain)	30 ml	750 ml	No drain	No drain	100 ml	No drain	No drain	
Hypotension during surgery	No	No	No	Yes, period with MAP 45	No	No	no	
Postoperative pain treatment	Epidural	Epidural	Epidural	Epidural	Epidural	Epidural	Epidural	
Postoperatuve neurological exam	Normal	Normal	Normal	Normal	Normal	Normal	Normal	

PF instrumented posterior fusion, *SPO* Smith–Petersen osteotomy, *MAP* mean arterial pressure

*No changes in neuromonitoring during screw placement and correction, however, in general, more variability in the signals throughout the case due to Friedreich's ataxia

related to the ICD10 codes for spinal deformities were reported and all unique patients < 18yrs where identified. The DHD data were stratified according the ICD10 codes for idiopathic scoliosis, syndromic scoliosis, neuromusculair scoliosis, congenital scoliosis and hyperkyphosis. Due to Dutch privacy regulations no information on subgroups with $n < 5$ could be provided, therefore, data were pooled in 2-year intervals in 2013/2014 and 2015/2016 to prevent missing data from the smaller diagnosis groups. Analysis of the DHD dataset revealed that the data of one of the non-academic hospitals was not included in the DHD database. The board of this hospital choose not to share their data with the DHD, whereas the ten other hospital did share their information. The data from the missing hospital could readily be obtained from their financial administration and was included in the study. As further validity evaluation of the dataset, the number of unique patients with pediatric spinal deformities operated in an academic hospital was compared between the national and the local dataset: a 1–2% variance was found.

Results

All hospitals responded to our request and all surgeons that encountered a case of delayed SCI completed a questionnaire for each identified case. A total of 10 potential cases with delayed SCI were identified in 7 of 11 Dutch scoliosis centres. Three were excluded: one because there was no clear documentation of a normal postoperative neurological examination, one because of intraoperative neuromonitoring events with normal neurological examination after surgery which deteriorated several hours after surgery and one because the event took place in 2011, outside the study period 2013–2019. A total of seven unique patients were included. These were diagnosed in 2017 ($n = 2$), 2018 ($n = 3$) and 2019 ($n = 2$) in six hospitals. Patient and deformity characteristics, description of the performed surgery, details about the SCI/recovery and surgical team are shown in Tables 1, 2, 3, 4. In 2013–2019, a total 2884 of pediatric deformity surgeries were performed in the Netherlands. The surgeons performed a median of 60 pediatric spine deformity surgeries per year (range 20–65) and most surgeons completed a spine fellowship (Table 4). The overall risk of delayed SCI in 2013–2019 was 1:412 surgeries (0.24%). With a risk of 1:201 (0.50%) and 1:214 (0.47%), delayed SCI was more common after neuromuscular and syndromic scoliosis surgery than in idiopathic scoliosis (1:595; 0.17%) (Table 5, Fig. 1).

Median age of the included patients was 16 yr. Median Cobb angle was 67°. Three patients with delayed SCI had adolescent idiopathic scoliosis, two patients had neuromuscular scoliosis (one kyphoscoliosis) and two patients syndromic scoliosis (Table 1). There was no case of delayed SCI identified after surgery for Scheuermann's kyphosis. All included patients had a primary surgery, no revision cases, median blood loss was 600 ml and median surgical time was 250 min. All patients had a documented normal postoperative neurological examination. Epidural catheters were used in the postoperative pain management of all reported cases (Table 2). Two patients had a complete SCI and five patient had an incomplete injury. Four patients were re-operated and

Table 3 Delayed SCI details

	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Median
Time between OR and diagnosis delayed SCI	7 h	16 h	32 h	40 h	6 h	36 h	2.5 h	16 h
ASIA score	ASIA B	ASIA A	ASIA B	ASIA C	ASIA B	ASIA A	ASIA B	
Level of SCI	C5-6	T7	T 10- 12	T3	T4-5	L 1	T7- 1 1	
Postoperative period with hypotension	Yes	Yes	Yes	Yes	Yes	Yes, after start epidural	Yes, after start epidural	
Hypotension during onset of neurological deficit	Yes	Unknown	No	No	No	No	Yes, after start epidural	
Hemoglobin levels at onset of deficit mmol/l	5.2	5.2	7.8	4.9	7.1	4.8	8.9	5.2
Time between diagnosis SCI and reoperation	43 h	12 h	3 h	–	13 h	–	–	12.5 h
ASIA score at most recent follow-up	ASIA-C	ASIA-C	ASIA B	ASIA A	ASIA-E	ASIA B	ASIA-E	
ASIA grade improvement (+) deterioration (-)	+1	+2	0	−2	+3	+1	+3	+1
Additional complications	Proximal and distal add on	–	Wound infection	Broken rod	–	–	–	
Hypothesis regarding cause of delayed SCI	Vascular, cervical proximal of scoliosis	Vascular, medial wall breach T6 screw	Hypotension	Traction and intra spinal epidural catheter	Vascular, medial wall breach T4 screw	Vascular	Vascular, hypotension after start epidural	

ASIA American spinal injury association score for SCI

underwent decompression and/or removal of spinal instrumentation. Five patients showed neurological improvement of their SCI, one patient showed no improvement and one patient deteriorated. All patients had a documented postoperative period with hypotension. Hypotension and vascular compromise of the spinal cord was the predominant hypothesis for the delayed SCI reported by the surgeons (Table 3).

Discussion

Delayed SCI is fortunately rare after pediatric spinal deformity surgery, but the risk found in this national cohort study was higher than previously reported. In this study, 7 cases (0.24%) of delayed SCI were identified among 11 hospitals treating 2884 patients in a 7-year study period. Most cases were incomplete and showed some neurological recovery during follow-up. Most surgeons reported delayed spinal cord ischemia as most likely cause of the delayed neurological deficit. The incidence of delayed SCI in neuromuscular and syndromic scoliosis was almost three times higher than in idiopathic scoliosis.

Few studies have described the risk of delayed neurological deficit after spine surgery, let alone delayed SCI after pediatric spinal deformity surgery. In 2016, Auerbach et al. conducted an online survey among active and emeritus surgeon members of the Scoliosis Research Society with an overall response rate of 38%. The estimated incidence of delayed SCI in the survey was 1 in 9910 cases (0.01%), which was almost 24 times lower than the risk identified in the current study. Of the reported cases, 69% were children and 50% consisted of unknown, radiculopathy and cauda equina symptoms [7]. Qiao et al. conducted a retrospective single center cohort study containing 5377 idiopathic scoliosis patients who underwent spinal deformity surgery [6]. They reported a total of seven cases of delayed

Table 4 Management delayed SCI

	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7
Elevation of MAP	Yes	No	Yes	Yes	Yes	No	Yes
Blood transfusion	Yes	Yes	No	Yes	No	Yes	No
Radiological evaluation	CT, CTA, MRI 2x	Yes, MRI	CT, MRI	CT, MRI	CT, MRI	CT, MRI	CT, MRI
Radiological findings	CT/CTA: T5 screw lateral breach Initial MRI: no findings 2nd MRI: after hardware removal: edema cranial of fusion trajectory (cervical)	MRI: possible compression medial breach T6 screw	CT: Hematoma T6-T10 MRI after decompression: SC edema	CT: No findings MRI 36 h after start deficit: SC edema T1-3. C6-T5 epidural swelling	CT: Medial breach T4 screw MRI:possible compression SC due to medial breach T4 screw	CT: No findings MRI: epidural lipomatosis	CT: T5 screw lateral breach MRI: no findings
Administration of corticosteroids/dexamamethasone	Yes after edema was seen on 2nd MRI	No	Yes	Yes	No	No	No
Reoperation	Yes 2x - Rod and T5 screw removal, cervical -Laminectomy C4-C6	Laminectomy T5-T6, T6 screw removal	Laminectomy T6-T10	No	Rod and screw removal and Laminectomy T4-T11	No	No
ASIA grade improvement (+) deterioration (−)	+1	+2	0	−2	+3	+1	+3

Table 5 Details regarding the surgical team

	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Median
Surgical team	2 surgeons, resident	1 surgeon, 1 resident	1 surgeon	1 surgeon	1 surgeon, 1 resident	2 surgeons	2 surgeons	
Dedicated spine anaesthesiologist	Yes	No	No	No	Yes	Yes	Yes	
Estimated annual number of scoliosis surgeries per surgeon	60	40	60	60	65	60	20	60
Years of experience with scoliosis surgery per surgeon	3 yr 3 yr	9 yr	28 yr	8 yr	15 yr	7 yr 20 yr	5 yr 10 yr	8.5 yr
Completed spine fellowship	Yes Yes	Yes	Yes	Yes	No	No No	Yes Yes	

neurological SCI, with three patients under 21 years of age. Their reported incidence of 0.10% is more comparable to the incidence for idiopathic scoliosis found the current study [6]. Unfortunately, patients with syndromic or neuromuscular scoliosis were not included.

There is a substantial difference between the 0.10 and 0.24% risk of delayed SCI reported by Qiao et al. and the current study and the 0.005% reported by Auerbach et al. [6, 7]. We can only speculate about the cause of this difference and we assume that it can be explained by the study setting as reflected by the low response rate: 38% versus 100%. The total number of deformity surgeries reported in the study by Qiao et al. and in the current study are obtained from an institutional and national database, respectively, whereas the total number of surgeries in the international study by Auerbach et al. is based on an estimation of surgeon

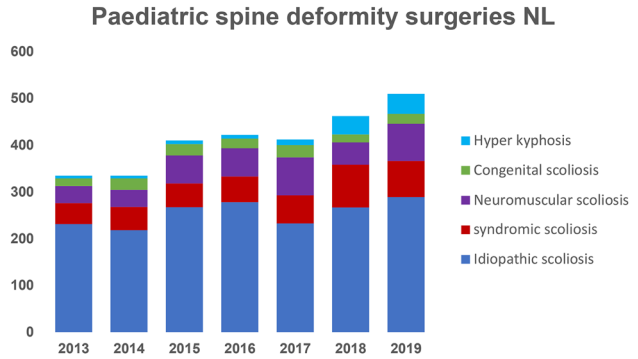

Fig. 1 Number of spine deformity surgeries in the Netherlands per year

reported volumes [6, 7]. Tsirikos et al. reported up to 21 spinal deformity surgeries in Scotland per 1,000,000 inhabitants per year [14]. In our national series, we found a similar incidence of 24 surgeries for scoliosis or kyphosis in children per 1,000,000 inhabitants per year. Therefore, the combination of a possible overestimation of the total number of performed surgeries and potential underestimation of the cases of delayed SCI due to the limited response rate of 38% could have led to the much lower risk reported by Auerbach et al. Moreover, the current study period is more recent 2013–2019 which could have led to a reduction in recall bias.

This is the first study reporting the risk of delayed SCI in syndromic and neuromuscular scoliosis, which appears to be almost three times higher than in idiopathic scoliosis. Indications for an increased risk for delayed SCI in syndromic syndromic and neuromuscular scoliosis can also be observed in two recently published case series [15, 16]. The case series by Alam et al. and Lovi et al. report a combined number of 22 cases of delayed SCI in which 64% of patients had a non-idiopathic scoliosis [15, 16]. The increased risk of complications in non-idiopathic scoliosis is well known and is mainly caused by underlying comorbidities [3]. Most likely, these comorbidities contribute to the increased risk of delayed SCI in non-idiopathic scoliosis surgery.

Since delayed SCI after pediatric spinal deformity surgery is rare, little is known regarding the optimal management of this devastating complication. As early identification of new neurological deficit allows for immediate action to be taken to identify the cause and potentially reverse the neurological changes, it is essential to serially monitor neurological function after pediatric spinal deformity surgery. Based on the anecdotal case reports in the current and previous studies, the optimal duration and frequency of postoperative monitoring cannot be specified. Basically, it is important to ensure that the personnel with direct care of the postoperative spinal deformity patient can recognize neurological changes in the first hours and days after surgery.

The exact etiology of delayed SCI is unknown but most studies suggest spinal cord hypoperfusion as most likely cause of delayed neurological deficit after pediatric spinal deformity surgery [15, 17]. The mechanical stretch on the spinal cord after deformity correction can lead to decreased perfusion of the spinal cord which, in combination with hypotension, could lead to ischemia of the spinal cord [7, 17]. Therefore, all factors that contribute to postoperative hypotension can potentially increase the risk of delayed SCI; e.g. peri-operative blood loss and epidural analgesia [7, 15, 17].

To date, there are not validated algorithms or postoperative checklists available to optimize the response to delayed SCI after pediatric spinal deformity surgery [18, 19]. Cessation of medication that could induce hypotension, maintain supraphysiological mean arterial blood pressure, acute MRI/CT to detect screw malposition or epidural hematoma, acute decompression or removal of spinal instrumentation are often suggested in literature [7, 17, 20]. Hemoglobin levels also effect spinal cord perfusion and elevated hemoglobin levels by red-cell transfusions have been associated with improved neurological outcome after traumatic spinal cord injury [21]. Although not described in literature, red-cell transfusion till at least physiological hemoglobin levels could be considered in the treatment of delayed SCI. In the present series, the treatment initiated was highly variable. In the majority of the cases, however, spinal imaging was performed before reoperation. We believe that a multi-disciplinary approach with an active communication between the surgeon, anesthesiologist, and intensive care physician is essential to review the events prior to the neurological deficit, review the peri-operative ischemic events and create attention to maintain adequate level of MAP and hemoglobin for adequate cord perfusion. When a reoperation is performed, it is crucial to maintain adequate MAP and hemoglobin levels during surgery to avoid a second hit of the SCI. Many facets of the the multi-disciplinary checklist for intraoperative neuromonitoring changes developed by Vitale et al. and the response algorithm by Lenke et al. are also useful in the treatment of delayed neurological deficit (Fig. 2) [18, 19].

The current study has some limitations. Delayed SCI is very rare complication and, therefore, we were only able to identify seven cases. Although this is sufficient to determine the incidence of this rare complication, seven cases is not enough perform any statistical analysis and we were, therefore, limited to descriptive statistics. More importantly, this study is retrospective and based on the recall of cases with delayed SCI by surgeons. Fortunately, pediatric spinal deformity surgery in the Netherlands is well organized and we had a 100% response rate on our questionnaires. Due to the use of electronic patient files, all case report forms could be completed without missing

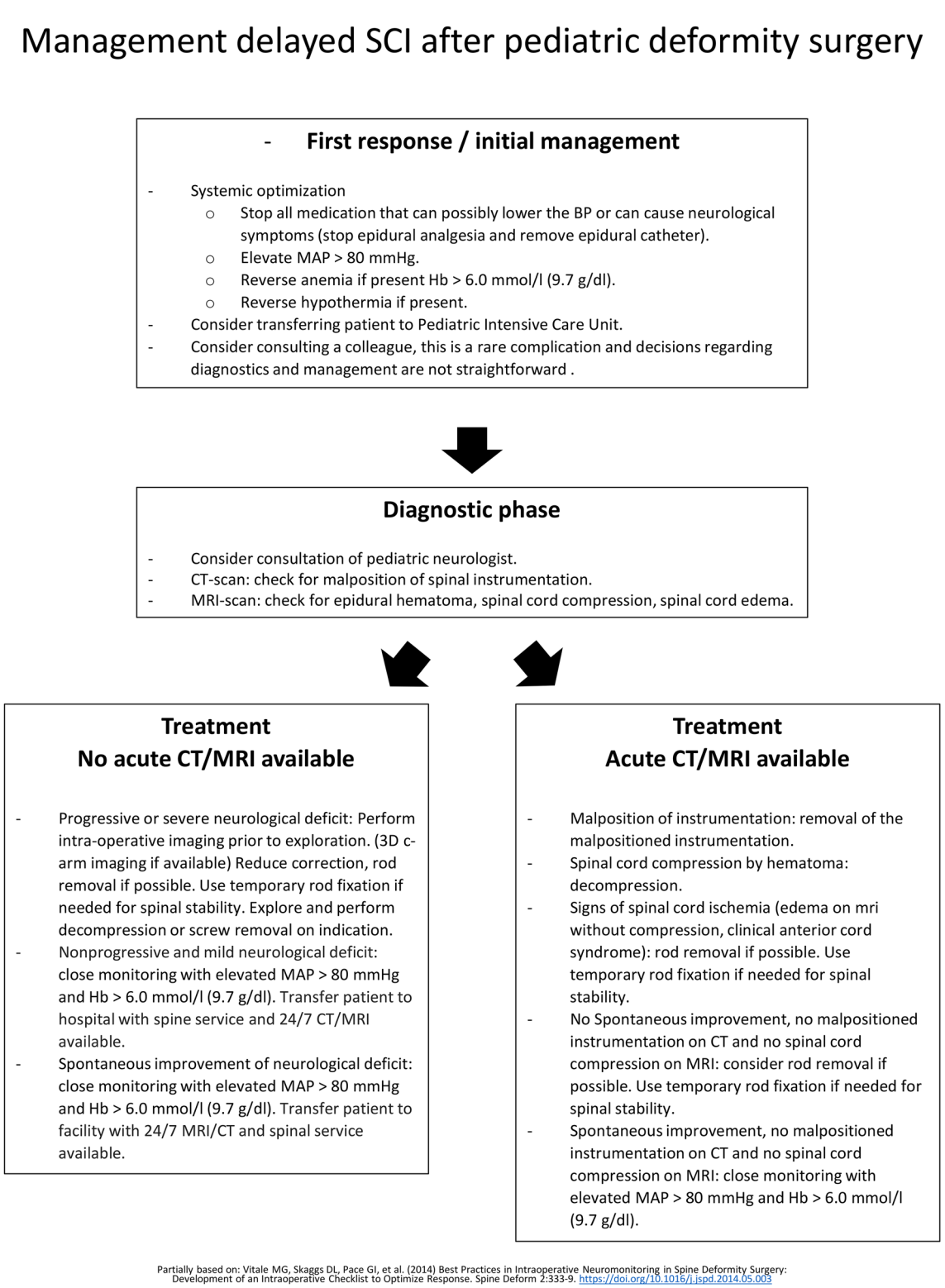

Fig. 2 Management delayed SCI after pediatric deformity surgery. *BP* blood pressure, *MAP* mean arterial pressure, *Hb* hemoglobin

data. Nevertheless, even if we would have missed one or more cases of delayed SCI, for example due to recall bias, this would only increase the incidence of delayed SCI and will only strengthen the conclusion of the current study. Therefore, the incidence of delayed SCI reported in the current study should be considered a minimum rate and the actual incidence of delayed SCI might even be higher.

Prospective registration of complications in large databases of national societies, governmental institutions or international scientific societies, such as the SRS Morbidity and Mortality database may help to better estimate the occurrence of this specific complication in the standard care, and may help to identify a larger sample to find ways to address this complication and improve patient outcomes in the future.

Conclusion

The current study demonstrated that the risk of delayed SCI after pediatric deformity surgery might be higher than previously assumed, especially in patients with non-idiopathic scoliosis. Since 90% of the delayed SCI occurs within 48 h after surgery [7], regular neurological follow-up during the first 2 days and nights after surgery is essential for timely diagnosis and management of this devastating complication.

Author contributions JPHJR, JJMR, DHRK, CF, AS, MCK, AM, PPH, LWLK, MK, RMC, TPCS: (1) made substantial contributions to the conception or design of the work; or the acquisition, analysis, or interpretation of data; or the creation of new software used in the work; (2) drafted the work or revised it critically for important intellectual content; (3) approved the version to be published; (4) agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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Data availability The datasets generated during and/or analysed during the current study are available from the corresponding author on reasonable request.

Declarations

Conflict of interest No conflict of interest for any of the authors.

Ethical approval This study was approved by the Medical ethical committee of the Erasmus MC: MEC-2022–0427.

Informed consent Informed consent was obtained from all included patients with delayed SCI.

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