



ARTICLE

Scoliosis in paediatric onset spinal cord injuries

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Abstract

Study design This is a retrospective longitudinal review.

Objective The purpose of this review was to identify predictors of developing clinical scoliosis and compare between traumatic and neurological aetiologies of SCI.

Setting This study was conducted at the Midland Centre of SCI.

Method Case notes of all patients injured at an age up to 18 years and admitted between 1971 and 2013 were reviewed.

Results Sixty-nine individuals were identified, of which seven were excluded: three with pre-existing scoliosis and four with spina bifida. The remaining 62 (44 males, 18 females) had a median age at injury of 17 years (inter quartile range 13–17). Of these, 51 (82%) had traumatic and 11 (18%) had neurological injury. Most (42/51; 82%) of the children who had a traumatic injury were older than 13 years. The risk of developing scoliosis was lower for older patients (RR 0.68 per year, 95% CI 0.52–0.83) or following a traumatic injury (RR 0.36, 95% CI 0.20–0.66). A multivariable analysis based on age and trauma showed that only older age decreased the risk. A robust Receiver Operator Curve analysis suggested 14.6 years as the optimal threshold to predict development of scoliosis within 10 years (Area Under the Curve; AUC 0.83 (95% CI 0.73–0.93), sensitivity 70% (95% CI 50–89%), specificity 89% (95% CI 74–100%).

Conclusion Our results suggest that age below 14.6 years was a predictor for scoliosis. Once adjustment is made for age, the incidence of scoliosis does not differ between traumatic and neurological aetiologies of paediatric SCI injury.

Introduction

Children and adolescents with spinal cord injury (SCI) are at risk of developing neuromuscular scoliosis which can impact their quality of life. In the UK the reported incidence of traumatic SCI is 15 per million [1] but the paediatric incidence is not known. The reported incidence of scoliosis among these patients is as high as 100% in the preadolescent and very young age group [2–6]. Age at the time of injury has been identified as a predictor for spinal curvature progression [3–5, 7, 8]. However, there is limited evidence about the scoliosis risk in SCI patients with

neurological aetiologies. Long-term follow-up is suggested to monitor for delayed development of spinal deformity in patients with operated spinal tumours [9] but no study has compared between traumatic and non-traumatic SCI. The Midland Centre of Spinal Cord Injuries (MCSI) is one of the 11 designated SCI centres in the UK and has managed spinal cord injuries patients of both traumatic and neurological aetiology from a wide catchment area including North Wales, Mid Wales, South Merseyside, Cheshire and the West Midlands. Most paediatric spinal injuries including ligamentous injury can be treated conservatively [10, 11]. However, given the alleged hazards of prolonged immobilisation in a child, especially with a halo, surgical stabilisation is the commonest treatment following traumatic injuries [12]. Standard care at the MCSI involves Active Physiological Management of traumatic injuries with at least 6 weeks of complete bed rest [13–15].

Even though scoliosis is a common complication of paediatric SCI, very little is known about predictive factors other than age or prevention. With this background we set out to perform a retrospective review of paediatric onset SCI and the risk of subsequent scoliosis.

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Aim

The aim of the review was to identify predictors of scoliosis in paediatric SCI patients and compare between traumatic and neurological aetiologies.

Method

The MCSI has a local database of patients, which was used to identify all patients admitted with paediatric (under 18 years) onset of injury. This review was a sub-study of large service evaluation exercise of long-term outcomes of paediatric SCI and was supported by the audit department at our hospital. Data sets were reviewed by the audit team and the study was not considered for ethical approval. Data collection for the service evaluation was done in November 2013. Eligible patients should at least be 10 years post the date of SCI. Neurologically intact patients were excluded from the search. Patients with preceding scoliosis were excluded from the analysis, as were patients with a congenital anomaly such as spina bifida, who were considered to have multifactorial risks (Fig. 1). Medical records were reviewed retrospectively and information about demographics, clinical condition and aetiology were obtained. Information comprising the basic data set as per International SCI data sets was obtained at three time points: time

of injury, 10 years post injury and the latest clinic assessment. Scoliosis was defined as curvature of the spine on sitting [16]. Radiological confirmation of scoliosis based on the Cobb angle was obtained for patients whose radiological records were available. All data generated or analysed during the study are included in the published article and its supplementary information files (Supplementary information Appendix 1).

Neurological evaluation and severity of injury was determined using the Frankel classification for grading of acute SCI [17]. If it was not possible to accurately determine Frankel grading on younger patients, the child was classified as having tetraplegia or paraplegia. The influence of categorical (gender, traumatic nature of the injury, Frankel category, type of paralysis) and continuous (age at injury and time to admission) independent variables on the risk of scoliosis was investigated using contingency table analysis (Chi-squared or Fisher exact test) and logistic regression, respectively. A multivariable logistic model, based on all predictors with univariable $p < 0.20$, was used to analyse if multiple variables could better predict scoliosis [18]. We considered interaction terms if coefficient values changed considerably from the univariable to the multivariable model. We converted odds ratios (ORs) to relative risks (RRs) using the methods suggested by Zhang and Yu and Grant [19, 20]. A receiver operator curve (ROC) analysis of age was performed to determine an ROC [21] curve and find an optimal threshold based on the maximum distance to the diagonal line (Youden criterion). In order to minimise bias in the cut-off point and its associated sensitivity and specificity, a robust bootstrap method based on 999 bootstrap samples was used.

The data were analysed using R vs 3.5.1 (The R Foundation for Statistical Computing, Vienna, Austria) using the packages epitools, sjstats, pROC and rcutpoint. A p value below 0.05 was assumed to denote statistical significance. Normally distributed data were summarised using medians and inter quartile ranges (IQR).

Results

Sixty-nine patients with paediatric onset SCI were identified, the first of whom was injured in August 1971. Seven patients, (three with pre-existing scoliosis and four with spina bifida, were excluded from our analysis (Fig. 1). The remaining 62 (44 males, 18 females) had a median age at injury of 17 years (IQR: 13–17). The median duration of follow-up was 28 years (IQR 22–33). The median time interval between onset of trauma and admission was 2 days (IQR: 1–12 days). The majority of patients (51/62; 82%) had a traumatic injury whereas 11/62 (18%) had a neurological aetiology. A road traffic accident was the

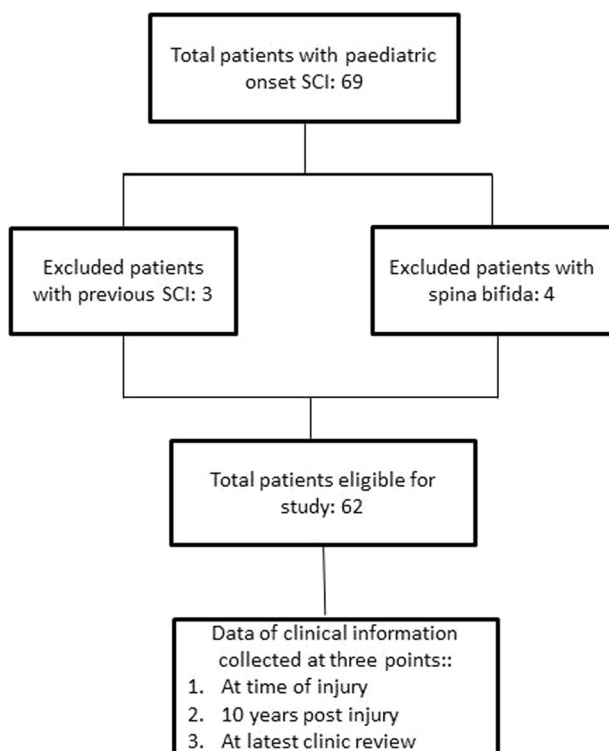


Fig. 1 Flow chart of the study group.

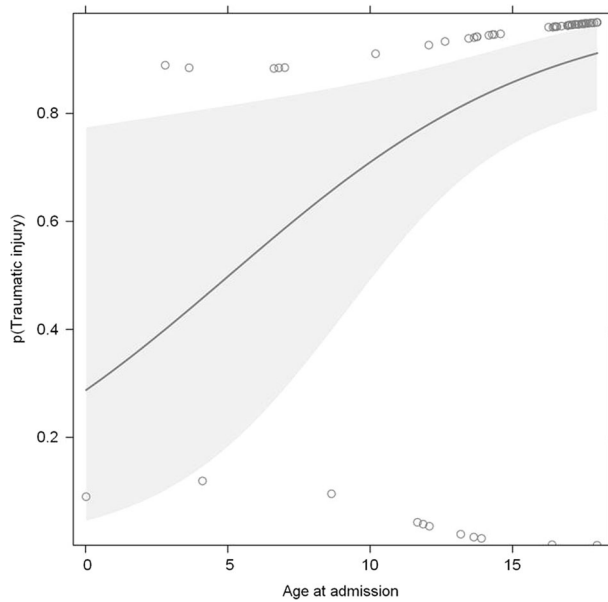


Fig. 2 Age as a predictor of admission with traumatic injury. Band represents 95% confidence intervals, and the vertical distances between open circles and predicted risk represent partial residuals (difference between predicted and actual risk).

commonest cause of trauma (37/51; 72%), with the remainder evenly split between a fall from height and a sports related injury (both 7/51; 14%). The neurological causes were evenly split between tumour (4/11; 36%), demyelination (4/11 (36%)) and vascular pathology (3/11; 28%). Older children were more likely admitted with trauma (RR 1.03/year, 95% CI 1.01–1.05, $p = 0.012$, logistic regression, Fig. 2). Adolescents aged 13 or above made up the vast majority of children who had a traumatic injury (42/51; 82%). There was no evidence that male gender predicted risk of trauma (RR 0.89, 95% CI 0.72–1.12, $p = 0.48$, Fisher's exact test).

Information of management of acute trauma was available for 48/51 (94%) of patients. Of these, 39/48 (81%) were managed conservatively, either by recumbence (31/39) or external immobilisation (8/39). The remaining 19% (9/48) of patients had surgical management of their spinal injury.

At the time of discharge, 4/62 patients (6%) had developed scoliosis, increasing to 19/62 (30%) 10 years post injury and 21/62 (34%) at the latest clinical assessment. For the majority of cases radiological data for scoliosis was not available because imaging was done at local hospitals and insufficiently reported on. The Cobb angle could be determined for seven patients, giving a median value of 70° (IQR: 21–72). Two further patients had confirmation of scoliosis based on MRI and another two based on abdominal X-rays.

The overall incidence of scoliosis was smaller in the traumatic group (13/52; 25%) than in the neurological group (8/11; 72%). Patients older at injury were less likely

Table 1 Influence of baseline characteristics on risk of developing scoliosis.

Baseline characteristic	10-year follow-up	
	Relative risk (95% CI)	<i>p</i> value
Univariable analyses		
Time to admission (per day)	0.99 (0.97–1.01)	0.46
Gender (male)	1.27 (0.55–2.9)	0.57
Age at injury (per year)	0.68 (0.52–0.83)	<0.001
Traumatic injury	0.36 (0.20–0.66)	0.01
Frankel grade	–	0.56
Tetraplegia	0.52 (0.18–1.5)	0.22
Multivariable analysis		
Age at injury (per year)	0.71 (0.54–0.86)	0.001
Traumatic injury	0.47 (0.08–1.6)	0.29

Continuous variables analysed using logistic regression; categorical and nominal variables analysed using Chi-squared (gender) or Fisher's exact (all others) test

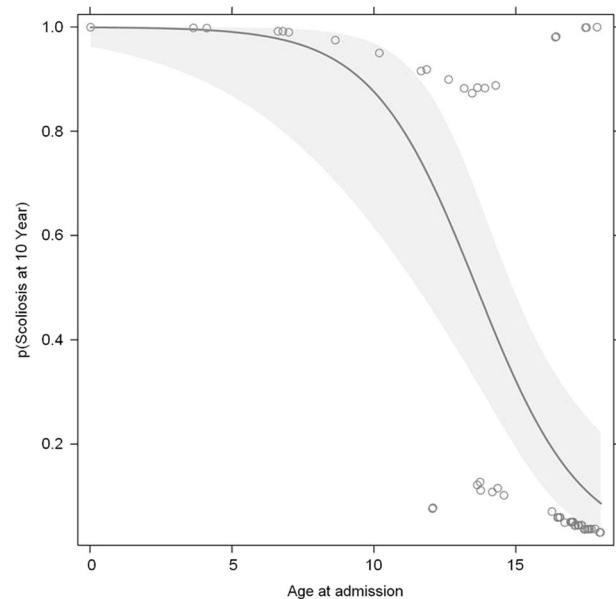


Fig. 3 Age as a predictor of developing scoliosis. The bands represent 95% confidence intervals, and the vertical distances between open circles and predicted risk represent partial residuals (difference between predicted and actual risk).

to have developed scoliosis at 10 years (RR = 0.68/year, $p < 0.001$; Table 1). No evidence was found that gender, level of injury, Frankel status or type of paraplegia predicted scoliosis onset (Table 1). A multivariable analysis based on age and trauma showed that younger age did ($p = 0.001$) but being non-traumatic did not ($p = 0.29$) predict development of scoliosis. According to the model, the predicted risk of scoliosis depended strongly on age and varied from near 100% for infants to around 10% for adolescents (Fig. 3). We therefore performed a robust ROC

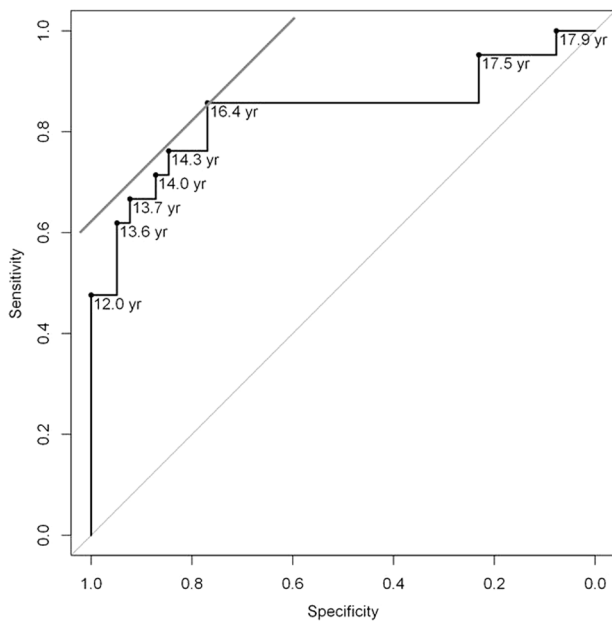


Fig. 4 ROC curve, showing sensitivity and specificity at various cut-off points when using age to identify patients at higher risk of developing scoliosis within 10 years from admission. The line represents Youden's criterion, suggesting several equivalent threshold ages. Using a robust bootstrapping method, an optimum threshold would be age below 14.6 years (AUC 0.83 sensitivity 70% and specificity 89%).

analysis for age among patients with a traumatic aetiology. Based on this analysis, an age of 14.1 years was the optimal threshold to identify patients admitted with trauma developing scoliosis within 10 years after admission (AUC (area under the curve) 0.78 (95% CI 59–93%), sensitivity 63% (95% CI 38–83%), specificity 91% (95% CI 79–100%)). Because of the small number of non-traumatic cases aged over 13, we repeated the ROC analysis for all patients. This suggested 14.6 years as the optimal threshold to identify any patient developing scoliosis within 10 years (AUC 0.83 (95% CI 0.73–0.93), sensitivity 70% (95% CI 50–89%), specificity 89% (95% CI 74–100%); Fig. 4).

Discussion

In our study, we found that the incidence of scoliosis was more than double amongst patients with non-traumatic aetiologies. However, we also found that age at injury and traumatic aetiology were correlated, with younger patients more likely to have a non-traumatic aetiology. Our multivariable analysis included both these predictors and found evidence that both were important, with one affecting the influence of the other. Specifically, age influenced the risk of scoliosis among patients admitted following trauma, but no evidence for an influence of age was found in patients admitted without trauma. An ROC analysis based on the

patients admitted following trauma found an age below 14.1 as the optimal cut-off point to identify patients at risk of developing scoliosis within 10 year. However, if one would disregard the interaction effect and only consider age, then an age below 14.6 years would be an optimal cut-off point, suggesting that in practice age alone is a good predictor.

Other studies also identified age at the time of injury as a predictor for spinal curvature progression [3–8, 20]. Mulcahey et al. [4] studied a group of children with an average follow-up of 4.2 years to analyse several risk factors of worse curvature and spinal fusion. They identified the American Spinal Injury Association Impairment Scale and age as the only two significant univariable predictors but their multivariable regression analysis left age alone as the best predictor. They used an age of 12 as an illustrative cut-off point for comparative risk calculations, but explain this age had no statistical value. Our study had a longer follow-up and found statistical evidence for the use of an age around 14 as an optimal cut-off point. No paediatric study has compared between traumatic and neurological aetiologies as a predictor for scoliosis.

Surgical management of trauma was not predictive for scoliosis in our study. It would be interesting to note if there is a change in this trend in the future as more paediatric patients are now surgically operated [12] to avoid the alleged risk of long-term immobilisation since the introduction of trauma networks in the UK from 2010. These are joint protocols for the management of traumatic SCI between each Major Trauma Network and affiliated Spinal Cord Injury Centre, mandated by the NHS Clinical Advisory Groups Report in 2011 [1].

This study has the typical limitations of a retrospective review, in particular a lack of robust radiological evidence of scoliosis and its progression. Instead, we used observation in sitting to define scoliosis. The risk factors and their associated RRs identified in this study therefore relate to the risk of observational scoliosis; their values may differ if a radiological definition for all our patients was available. A prospective study would also have recorded the time point of scoliosis diagnosis. With that information, we could have performed a more powerful time-to-event (survival) analysis and could perhaps better analyse if the time since admission would predict the risk of scoliosis. Strategies like use of standing frame or compliance with the TLSO which can have a mitigating effect on development of scoliosis cannot be readily quantified nor were they evaluated. This study obtained cross-sectional information at three time points and therefore continuous data for spinal progression were not available. Despite these limitations the authors have complied with the STROBE checklist for observational studies.

The findings of this study may have implications on the management and the rehabilitation programmes for children

with SCI by identifying predictors for scoliosis and supporting long-term follow-up. The findings may also have a role in enhancing the understanding about evolution of scoliosis in paediatric SCI.

Data availability

Data supporting the results can be obtained from the corresponding author.

Author contributions RK: oversaw the design of the study, wrote the manuscript, did data collection and contributed to the analysis and interpretation of results. JK: performed statistical analysis and edited the main article. WEIM: contributed to revising the manuscript and data interpretation. JC: contributed to study design, methodology and data interpretation. SK: helped with radiology data collection. NK: contributed to study design, methodology and data interpretation. RL: helped with radiology data analysis. AO: contributed to study design, methodology and data interpretation.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval Healthcare quality improvement partnership (HQIP) provides guidance intended to help those responsible to review and develop arrangements for the effective ethics oversight of quality improvement and clinical audit activities, as required. Our local audit department follows the principles of HQIP. This study was approved by the local audit department allowing use of clinical data for service evaluation (Supplementary Appendix 2, certificate from audit department).

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