



Predictors for spinal deformity following resection of intramedullary tumor via posterior approach: a systematic review and meta-analysis

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Abstract

Purpose The present study aimed to identify the clinical predictive factors for worsened spinal deformity (SD) following surgical resection via posterior approach for primary intramedullary tumors.

Methods A systematic search was performed using PubMed, Web of Science, and Scopus databases to extract potential references. Observational studies reporting predictive factors for worsened SD following surgical resection via posterior approach for primary intramedullary tumors were included. The odds ratio (OR) was calculated for dichotomous parameters.

Results Four retrospective cohort studies were included in the meta-analysis. They were comprised of two groups of patients; those who developed SD ($n=87$) and those who did not ($n=227$). For patients with IMSCTs, age under 25 years as well as age under 13 years were the demographic variables associated with postoperative SD (odds ratio [OR] 3.92; $p=0.0002$ and OR 4.22; $p=0.003$). In both the fusion and the non-fusion subgroups, preoperative spinal deformity strongly predicted postoperative SD (OR 11.94; $p<0.001$), with the risk highly elevated among the non-fusion patients (OR 24.64; $p<0.0002$). Thoracolumbar junction involvement was also found to be a predictor of postoperative SD for patients with IMSCT (OR 2.89; $p=0.02$).

Conclusion This study highlights the importance of considering age, preoperative spinal deformity, and thoracolumbar junction involvement as predictors of postoperative spinal deformity following surgical resection for IMSCT. These findings may provide guidance for the management of these patients, including the development of preoperative planning strategies and the selection of the most appropriate surgical approach for high-risk patients.

Keywords Spinal deformity · Resection · Intramedullary spinal cord tumor · Laminectomy · Laminoplasty

Introduction

Intramedullary spinal cord tumors (IMSCTs) are rare forms of central nervous system (CNS) neoplasms with a prevalence as low as 2–10% of all CNS neoplasms and an incidence of less than 1/100,000 people per year [1–4]. It most commonly presents with symptoms such as pain, sensory/motor deficits, and sphincter dysfunction [3]. Eventually,

the majority of them require surgical resection. The access to the spinal canal is usually obtained by laminectomy, laminoplasty, or laminotomy, referred to as “posterior approach” methods.

Employing laminectomy as a surgical approach, accessibility to the spinal canal is accomplished by detaching the paraspinal muscles. This is followed by the removal of the spinous processes and associated ligaments, including the posterior interspinous ligament, which is a key component of the tension band mechanism [2]. Consequently, this detachment and removal leads to a reduction in the force exerted by the extensor muscles. As demonstrated by Pal et al. [5], the posterior columns are responsible for transmitting as much as 64% of the axial load.

Due to the decrease in extensor force following the procedure, there is a shift in load toward the anterior aspect of the spine. This shift, in a significant number of patients,

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culminates in postoperative spinal deformities (SD), predominantly kyphosis [6]. The growing spine is considerably more susceptible to deformity; thus, children and young adults exhibit a higher risk of developing SD post-laminectomy [7]. Reported incidence rates show substantial variability. Some studies have documented incidence rates as high as 64% among the younger population [8]. It is essential to interpret these figures with caution, especially when considering differences in surgical procedures and relatively low number of analyzed cases.

Laminoplasty, in which the posterior elements of the spinal column are replaced following *en bloc* removal of the laminae, has been introduced to avoid the complications seen in laminectomy. However, its effectiveness in preventing postoperative kyphosis is yet to be demonstrated [4, 9–11]. The purpose of this study is to determine the clinical predictive factors for worsened spinal alignment following surgical resection of IMSTs via posterior approach, in order to indicate whether delayed spinal fusion may be required to prevent postoperative spinal deformity.

Methods

Search strategy

The screening procedure adhered to the guidelines outlined in the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA). A comprehensive literature search was conducted utilizing PubMed, Web of Science, and Scopus databases from the earliest available records until October 22, 2022, including studies published in all languages. The search was conducted using the following keywords: ((laminoplasty) OR (laminotomy) OR (laminectomy)) AND ((tumor*) OR (resection)) AND ((spinal_deformity) OR (kypho*)). The search was performed independently by one author (M.S.). The study's protocol was registered through PROSPERO (registration no. CRD42023377675; <https://www.crd.york.ac.uk/prospero/>).

Selection criteria

The initial phase of screening involved a comprehensive evaluation of the titles and abstracts of the articles, in order to ascertain that they met three predefined criteria. These were: (1) written in the English language, (2) involving patients undergoing surgery via posterior approach for the treatment of IMSTs, and (3) reporting risk factors for postoperative SD. The articles that passed the initial screening were then subjected to a secondary screening phase. This phase assessed whether the articles provided sufficient extractable data, and if they met the minimum sample size requirement (i.e., at least 10 patients). Any articles that

failed to meet either of these criteria were excluded from further analysis. Screening was performed independently by two authors: M.S. and J.K. Disagreement at any step was resolved by the author, G.M.

Quality and publication bias evaluation

The quality of each study selected for analysis was evaluated using the modified Newcastle–Ottawa Scale (NOS). A study could earn a maximum of 2 points in the comparability category and a maximum of 1 point in the other categories, for a total of 9 points. A score of 7 or higher indicated high quality, a score between 6 and 4 indicated moderate quality, and a score of 3 or lower indicated low quality. The quality assessment was performed independently by one author (O.L.), and any disagreements were resolved through discussion with author G.M. Publication bias among the included studies was assessed through visual inspection of funnel plot asymmetry.

Statistical analysis

For the statistical analysis, dichotomized variables such as age and sex were pooled into an overall odds ratio (OR) and 95% confidence intervals (CI) to identify risk factors for postoperative SD. Additionally, patients were divided into subgroups depending on whether their diagnosis required fusion to assess any differences between the two groups. Random-effect models were used, and the heterogeneity of the overall OR was calculated using the I^2 statistic. Publication bias was again assessed through visual inspection of the funnel plot asymmetry. A p -value < 0.05 was considered statistically significant. All analyses were performed using Review Manager version 5.4.1 (Cochrane IMS).

Results

Search results

The comprehensive literature search resulted in the identification of 2730 unique records. The titles and abstracts of these records were critically evaluated for relevance, and 2697 records were subsequently excluded. The remaining thirty-three records underwent further analysis to evaluate the presence of extractable data. Twenty-nine records were determined to have insufficient data and were excluded from further consideration (Fig. 1). Hence, the final number of records included in this meta-analysis was four: they are listed in Table 1. They consisted of retrospective cohort studies, all rated as high quality according to the NOS (Supplementary Table 1).

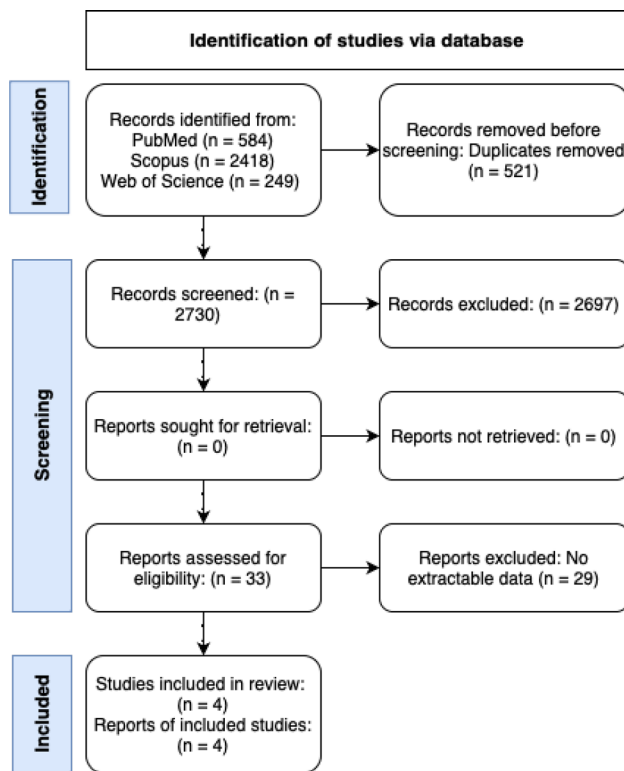


Fig. 1 PRISMA flowchart of the scientific literature search and study selection. Data added to the PRISMA template (from Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *BMJ*. 2021; 372:n71) under the terms of the Creative Commons Attribution License. Figure is available in color online only

Publication bias

The funnel plot revealed an asymmetrical distribution of the included studies in the graph between the mean deviation (MD) and standard error, indicating the presence of publication bias (Supplementary Fig. 1).

Predictors of spinal deformity

A total of 20 extractable perioperative variables reported in the literature were assessed (Figs. 2, 3, 4, and 5, Supplementary Figs. 2–17). The identified statistically significant predictors for postoperative SD consisted of two demographic factors, one preoperative factor, and one intraoperative factor. For patients with IMSCT, age under 25 years was a demographic variable associated with postoperative SD (OR 3.83, 95%CI 1.59–9.22; $p = 0.003$) (Fig. 2). Additionally, age under 13 years was also displayed as a predictor for the development of spinal deformity post-tumor resection (OR 4.22, 95%CI 1.61–11.06; $p = 0.003$) (Fig. 3).

Table 1 Characteristics of included studies

Study	Country	Level of quality	Study design	Mean age in years	Definition for SD	Date of SD diagnosis	Pathology	Number of pts		Total
								SD	No SD	
Yeh et al. [15]	UK	High	Retrospective	5.57 ± 3.92	NR	NR	IMSCT	12	15	27
Montano et al. [17]	Italy	High	Retrospective	47.28 ± 14.77	ΔCA > 10°	NR	IMSCT	5	16	21
Shi et al. [16]	USA	High	Retrospective	37.0 ± 14.5	ΔCA > 10°	NR	IMSCT	27	78	105
Yao et al. [8]	Israel	High	Retrospective	8.6 ± 5.7	NR	NR	IMSCT	43	117	161

SD Spinal deformity; NR Not reported; IMSCT Intramedullary spinal cord tumor; and pt Patient

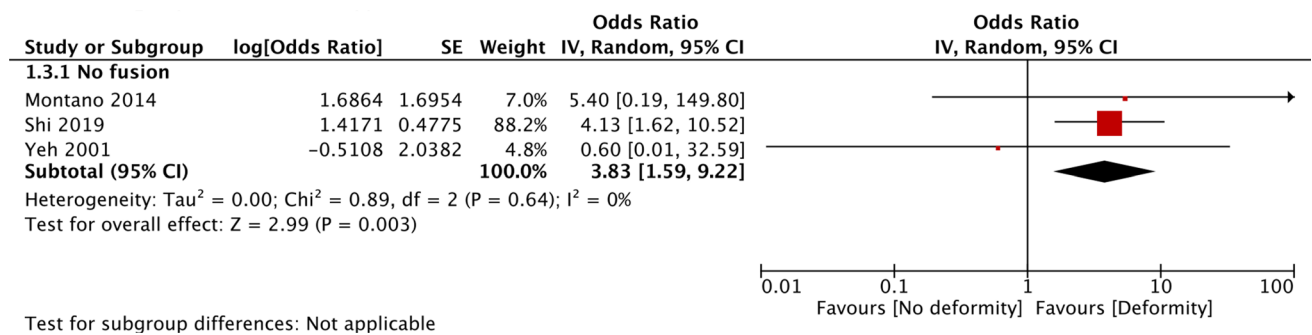


Fig. 2 Forest plot comparing the difference in risk for SD among patients 25 years old or younger with patients older than 25 years old. IV = inverse variance

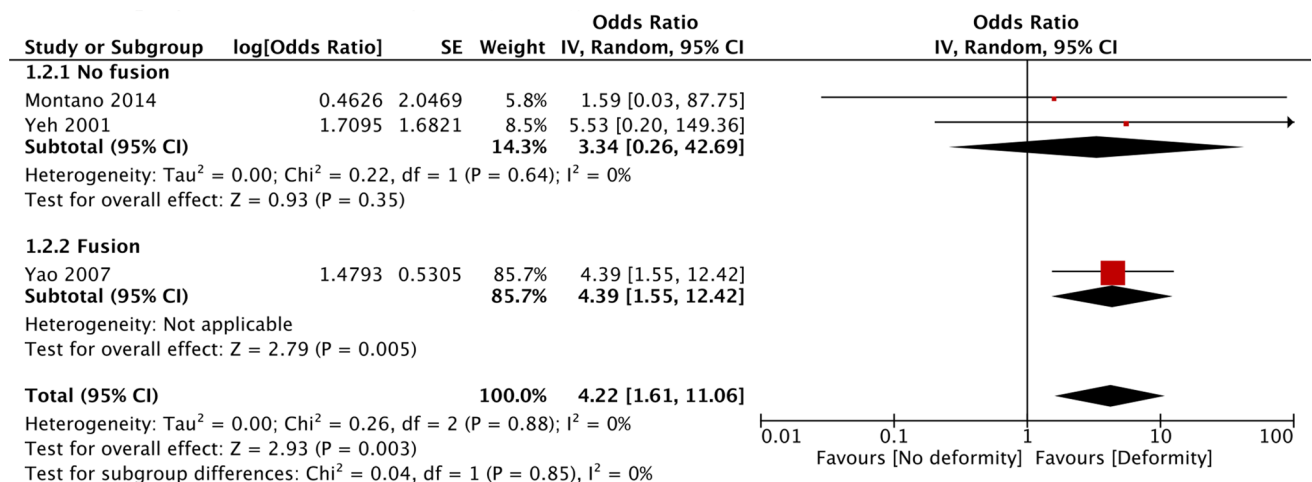


Fig. 3 Forest plot comparing the difference in risk for SD among patients younger than 13 years old with patients 13 years old or older

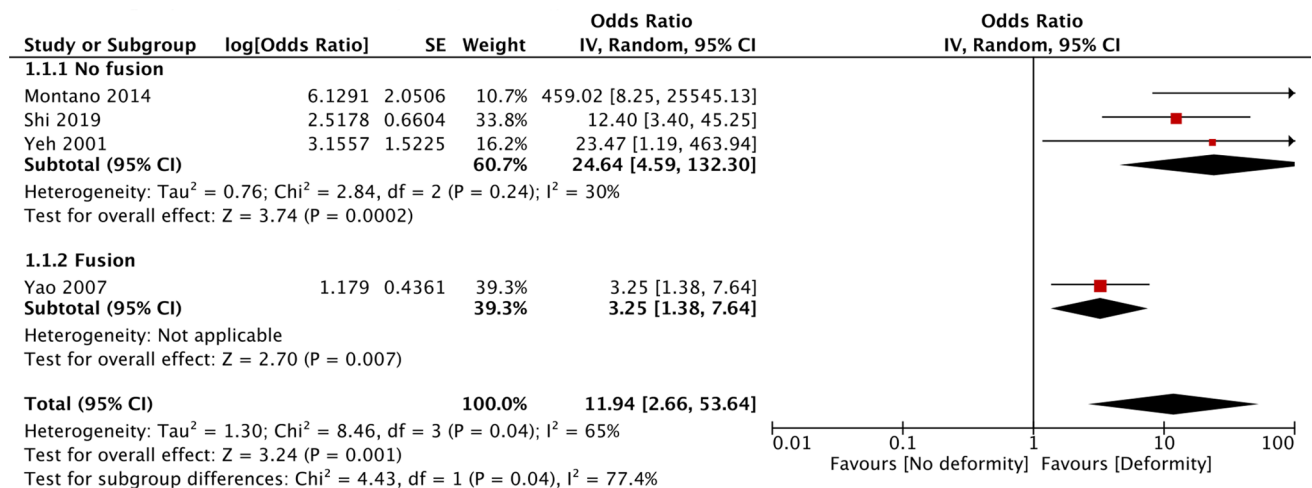


Fig. 4 Forest plot comparing the difference in risk for SD among patients suffering from preoperative SD with patients without preoperative SD

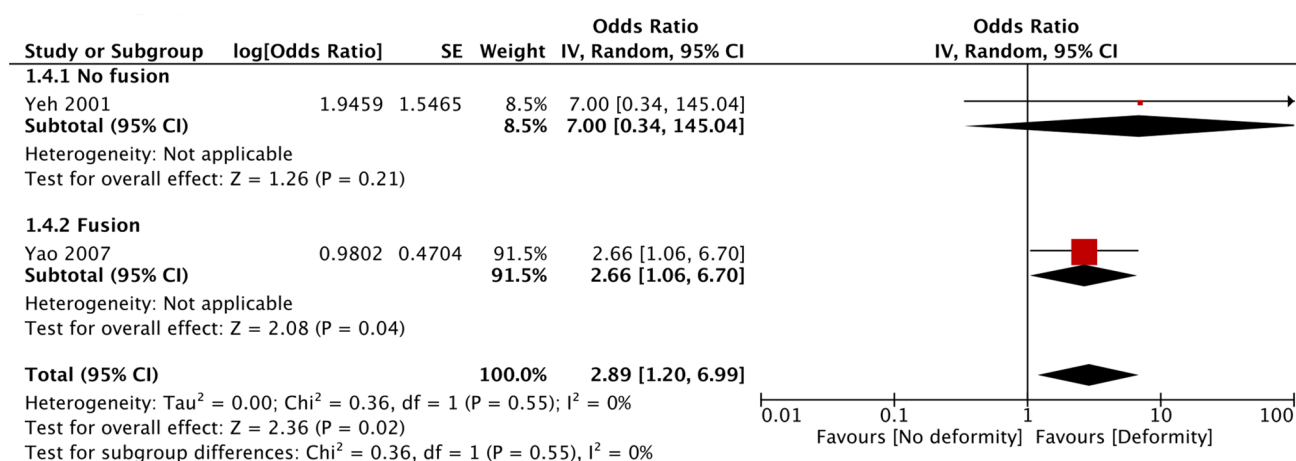


Fig. 5 Forest plot comparing the difference in risk for SD among patients undergoing surgery involving thoracolumbar junction with patients undergoing surgery excluding thoracolumbar junction

When pooling the fusion and non-fusion subgroups, the presence of preoperative SD was shown to be a strong predictor for postoperative SD (OR 11.94, 95%CI 2.66–53.64; $p < 0.001$) (Fig. 4). Furthermore, when assessing only non-fusion patients, preoperative spinal deformity was a statistically significant predictor for worsened SD (OR 24.64, 95%CI 4.59–132.3; $p < 0.0002$) (Fig. 4).

Thoracolumbar junction involvement and non-thoracolumbar junction involvement were assessed as predictors for postoperative SD. When assessing all patients, the thoracolumbar junction involvement group had a statistically significant increased prediction for SD (OR 2.89, 95%CI 1.20–6.99; $p < 0.02$) when compared to the latter (Fig. 5).

Variables that are lacking association with spinal deformity

All perioperative variables reported among the included studies were assessed for association with postoperative SD. Among the 20 variables, the following 16 were not statistically significant: sex, MMS at follow-up, preoperative MMS, astrocytoma, ependymoma, cervical involvement, thoracic involvement, lumbar involvement, cervicothoracic involvement, tumor confined in cervical region, tumor confined in thoracic region, tumor confined in lumbar region, single-level approach, two-level approach, 2 + level approach, laminoplasty, or laminotomy (Supplementary Figs. 2–17).

Discussion

This systematic review revealed that spinal alignment and age emerged as pivotal predictors in the progression of spinal deformity. The implications are profound. Additionally, regardless of whether patients underwent fusion

or not, pre-existing SD was a dominant predictor of worsened postoperative outcomes, with an even higher odds ratio observed exclusively in non-fusion patients. Given that many neurosurgeons working on intradural intramedullary and extramedullary tumors are more entrenched in brain surgery than spine surgery, there is a pressing need for heightened awareness, as emphasized by this study, about spinal deformity and its predictive factors among this group of practitioners.

Of the four retrospective studies included, a total of 314 patients were included in this analysis. To the best of our knowledge, this is the first systematic review and meta-analysis to identify predictors for SD following IMSCT resection.

Age

Our study consists of patients of all ages, but is mainly focused on two age groups: patients under 25 years old and those under 13 years old. We found that both groups demonstrated a statistically significant prediction for spinal deformity ($p = 0.003$). It suggests that younger individuals, whose skeletal systems are still developing, may have an increased risk of developing spinal deformity post-resection [7]. The higher incidence of spinal deformity in younger populations after IMSCT resection may be attributed to a combination of factors, including age-related differences in spine elasticity, stress concentration in key areas, and progressive elastoplastic strain. In adults, deformation mainly impacts viscous–elastic structures, leading to motion segment instability and vertebral remodeling [12, 13]. Further research should, therefore, focus on finding the optimal cutoff age that accurately predicts postoperative SD, as this has yet to be identified [14, 15].

Preoperative spinal deformity

The previous studies have shown that preoperative spinal deformity tends to worsen postoperative spinal deformity ($p=0.07$) [8, 15–17]. When pooling data together in this systematic review, we find compelling evidence that preoperative spinal deformity does predict postoperative spinal deformity ($p=0.001$). Therefore, it seems to be an important factor to take into account when choosing treatment for patients with IMSCTs. Advances in preoperative planning and the use of advanced MRI might reduce the risk of postoperative spinal deformity when the patient is displaying preoperative deformity [18].

Preoperative spinal deformity has been frequently described throughout the literature in individuals receiving IMSCT resection. In their analysis of 55 pediatric IMSCT patients, Ahmed et al. found that an underlying abnormality was present in 20% of cases [19]. A preoperative abnormality was present in 56 out of 161 pediatric IMSCT patients, and 46% of those deformities progressed, 43% remained stable, and 11% resolved postoperatively [8]. It has been cogitated that involvement of the anterior horn of the spinal cord during IMSCT may cause muscular weakening. This is caused by diminished innervation of nearby muscles, which could result in an increased spinal deformity [15, 20].

Thoracolumbar junction involvement

When examining the relationship between thoracolumbar junction involvement and spinal deformity, a statistical significant finding emerges. The odds of predicting spinal deformity in this group of patients are present ($p=0.02$). We can speculate that this finding appears due to the involvement and the ramification of the thoracic column and its osseoligamentous structures. When the posterior osseoligamentous structures are left intact, local denervation of the back muscles does not ultimately lead to deformity of the spine. This explanation was discovered and explored using an immature animal model [21].

The unparalleled incidence of spinal deformity explored in this study is not unique for IMSCT and has been observed in patients undergoing surgery in the thoracolumbar areas for trauma-related injuries. These patients have shown an increased incidence of postoperative spinal deformity [22]. It has been postulated that due to the high-stress nature of this region, which experiences rotational forces from the thoracic spine and significant mobilization stimulus from the lumbar region, spinal deformity tends to manifest in this area [23].

Limitations

This meta-analysis presents several limitations. Only one of the four studies presented preoperative Cobb angle

measurements, and only two of the four provided the definition of the degree in which the Cobb angle needed to change before being considered significant. Instead, the definition was categorized as either “worse” or “not worse” and “requiring fusion” or “asymptomatic.” Due to this, despite being scored as high in quality, the studies included in this meta-analysis have lower reliability. Of the 20 variables assessed for association with spinal deformity (SD), only four were found to be significant. However, it is important to note that the lack of significant association for some variables may be due to limited evidence. Variables such as tumor type and locations in the spine should not be disregarded, as they require further assessment in the future studies (p values close to significant and/or lacking studies assessing the variable). Furthermore, different studies reported on a limited number of the same variables, which could result in different outcomes observed by different groups of surgeons. Given that IMSCT is a rare type of pathology, our study only contains a total of 314 patients, which might make it challenging to draw large conclusions about the population as a whole. Since a multivariate analysis was not possible to perform, this study does not provide independent predictors for SD after surgical resection. To account for heterogeneity, a random-effect model was used for meta-analysis. However, this method may not account for all heterogeneity. Owing to the highly specialized nature of the subject matter, it is acknowledged that certain cited references may appear antiquated. Despite diligent efforts to identify more contemporary research, such endeavors have been unsuccessful in yielding up-to-date sources.

Conclusion

In conclusion, this systematic review and meta-analysis aimed to evaluate the available literature on preoperative, postoperative, and intraoperative variables that may be associated with SD following surgical resection of IMSCTs. Our analysis revealed that following key variables: lower patient age, pre-existing spinal deformity, and thoracolumbar involvement were statistically significant predictors of postoperative SD.

Further research is necessary to develop a clinically applicable preoperative risk scoring system which takes into account the identified cutoff points of the significant predictors. While the findings from this study should be interpreted with caution, the variables identified may be of great value to those seeking to develop a preoperative scoring system for risk analysis to predict SD in the clinical setting [24].

Supplementary Information The online version contains supplementary material available at <https://doi.org/10.1007/s00586-023-07957-1>.

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Declarations

Conflict of interest None reported.

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