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Int J Spine Surg published online 5 October 2023
https://www.ijssurgery.com/content/early/2023/10/03/8518

The International Journal of Spine Surgery
2397 Waterbury Circle, Suite 1,
Aurora, IL 60504, Phone: +1-630-375-1432

International Journal of Spine Surgery, Vol. 00, No. 0, 2023, pp. 1–9
https://doi.org/10.14444/8518

Clinical and Economic Impact of Proximal Junctional Kyphosis on Pediatric and Adult Spinal Deformity Patients

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ABSTRACT

The common goal of pediatric and adult spinal reconstructive procedures is to minimize long-term risk of disability, pain, and mortality. A common complication that has proved particularly problematic in the adult spinal deformity population and that has been an area of increased research and clinical focus is proximal junctional kyphosis (PJK). The incidence of PJK ranges from 10%–40% based on criteria used to define the condition. Clinically, PJK complication is associated with increased pain, decreased self-image and Scoliosis Research Society scores, and severe neurological injuries affecting the patient's quality of life. Economically, direct costs of PJK complication-associated revision surgery ranges from $20,000 to $120,000, which places an enormous burden on patients, providers, and payers. To mitigate the risk of PJK occurrence postoperatively, it is paramount to develop consistent guidelines in defining and classifying PJK in addition to extensive preoperative planning and risk stratification that is patient specific. This article will provide an overview on the clinical and economic impact of PJK in pediatric and adult spine deformity patients with an emphasis on the role of patient factors and predictive analytics, challenges in developing a consistent PJK classification, and current treatment and prevention strategies.

Other and Special Categories

Keywords: ASD, complication, cost, economics, PJK/PJF, PROMs, outcomes

INTRODUCTION

Examining the clinical and economic impact of proximal junctional kyphosis (PJK) on pediatric and adult spinal deformity (ASD) patients demands appreciation of the wide spectrum of spinal conditions and patient populations included in this review. The common goal of pediatric and adult spinal reconstructive procedures is to minimize long-term risk of disability, pain, and mortality associated with progressive spinal deformity. Evolution in the surgical treatment of complex spinal disorders benefited from tremendous advancement in spinal implants, osteobiologics, surgical techniques, and more specific classification of spinal deformity pathology in the period from the mid-1980s to early 2000s. Over the past decade, there has been an increased emphasis on outcome-based research and value-based care.

Given that surgical treatment of pediatric spinal deformity and ASD is associated with high index surgical costs, optimization of cost-effectiveness can only occur over a long-term time horizon and will require increased durability of procedural intervention with reduction of reoperations. One of the complications that has proved particularly problematic in the ASD population and that has been an area of increased research and clinical focus is PJK.

PJK DEFINITIONS, CLASSIFICATIONS, AND EPIDEMIOLOGY

One of the challenges in summarizing the clinical and economic impact of PJK on surgically treated ASD patients is the lack of a consistent definition and classification. Lowe and Kasten reported on the prevalence of PJK in patients with Scheuermann kyphosis who had undergone spinal fusion using Cotrel-Dubousset instrumentation.[1] Their report highlighted potential seriousness of the complication and the hazards associated with overcorrection of sagittal plane deformity. Lee et al found a 46% prevalence of proximal kyphosis at 2-year follow-up after hook and rod instrumentation in adolescent idiopathic scoliosis (AIS), defining abnormal kyphosis from T2 to the proximal level of the instrumented fusion as kyphosis of more than 5° above the summed normal angular segments.[2]

Glattes et al reported a 26% incidence of PJK in ASD patients and defined PJK with the criteria of both (1) a proximal junctional angle (PJA) of greater than or equal to 10° and (2) a PJA greater than 10° compared with

Table 1. Classification of the grades and severity of proximal junctional kyphosis and PJF.[10]

Classification	Description
Type	
1	Disc and ligamentous failure
2	Bone failure
3	Implant/bone interface failure
Grade	
A	Proximal junctional increase 10°–19°
B	Proximal junctional increase 20°–29°
C	Proximal junctional increase 30°
Spondylolisthesis	
PJF-N	No obvious spondylolisthesis above UIV
PJF-S	Spondylolisthesis above UIV

Abbreviations: PJF, proximal junctional failure; UIV, uppermost instrumented vertebra.

Table 2. The Hart-International Spine Study Group proximal junctional kyphosis severity scale.

Parameter and Qualifier	Severity Scale
Neurological deficit	
None	0
Radicular pain	2
Myelopathy/motor deficit	4
Focal pain	
None	0
VAS ≤ 4	1
VAS ≥ 5	3
Instrumentation problem	
None	0
Partial fixation loss	1
Prominence	1
Complete fixation loss	2
Change in kyphosis/PLC integrity	
0°–10°	0
10°–20°	1
>20°	2
PLC failure	2
UIV/UIV+1 fracture	
None	0
Compression fracture	1
Burst/chance fracture	2
Translation	3
Level of UIV	
Thoracolumbar junction	0
Upper thoracic spine	1

Abbreviations: PLC, posterior ligamentous complex;UIV, uppermost instrumented vertebra; VAS, visual analog scale.
Note: Reprinted with permission from Hart et al.[13]

preoperative measurements.[3] The angle was measured from the caudal end plate of the upper-instrumented vertebrae (UIV) to the cephalad end plate of the vertebral body 2 levels above the UIV, and 10° was selected largely based on the reliability of radiographic measurements,[4] without a firm anchorage to clinically significant PJK. Helgeson et al proposed a critical angle of 15° across one segment to define PJK based on the SD of all postoperative patients in their study group and the assumption that normal is within 2 SDs. They classified the patients into 4 groups based on construct type and reported an 8.1% PJK rate as their highest subgroup incidence rate using this more stringent definition.[5] Both methods for measuring the PJA (using UIV+1 or UIV+2) have demonstrated adequate reproducibility.[6] Bridwell et al also reported an angle of ≥20° as a possible critical angle to define PJK in ASD and reported a PJK rate of 27.8% at 3.5 years postoperatively.[7]

Wide variation in the incidence of PJK in various spinal deformity patient populations as well as a lack of clarity regarding its true frequency as a clinically problematic complication prompted efforts for more detailed classification systems.[8] The Boachie-Adjei classification[9,10] included grades and severity (Table 1).

To help better correlate PJK classification with validated patient-reported outcomes (PRO), the Hart-ISSG Proximal Junctional Severity Scale (Hart-ISSG PJKSS) was later developed (Table 2). This system evaluates the severity of neurological deficit, focal pain, instrumentation problems, change in kyphosis/posterior ligament complex integrity, UIV/UIV+1 fracture, and the level of the UIV in the setting of PJK and is a reliable and repeatable classification system for assessing patients with PJF, with higher PJF severity score scales correlate with recommendation for operative revision.[11] Lau et al[12] verified this scale to be correlated with validated patient reported outcome measures (PROMs): Oswestry Disability Index, visual analog scale pain, and the Scoliosis Research Society 30 (SRS-30) questionnaire.

PROXIMAL JUNCTIONAL FAILURE

The term proximal junctional failure (PJF) has been used to describe a more severe subset of PJK that is associated with adverse effects on patient outcomes and likely need for revision surgery (Figure 1). Hart et al described PJF as an increase in the PJA greater than 10° combined with one or more of the following: fracture of the vertebral body of UIV or UIV+1, posterior osseoligamentous disruption, or pullout of instrumentation at UIV.[13] Hostin et al proposed defining PJF occurring within 28 weeks of index surgery and resulting any of the following: a 15° increase postoperative PJA, vertebral fracture of UIV or UIV +1, failure of UIV fixation, or proximal extension of the fusion.[14] This study by Hostin et al, which included a total of 1218 consecutive ASD surgeries across 10 deformity centers, identified a PJF rate of 5.6%. Using the same definition, Annis et al reported a PJF incidence of 38.5% for their retrospective review of 135 consecutive patients with a minimum 2-year follow-up, treated at a single institution for ASD with a UIV in the thoracolumbar spine (T9–L2).[15] Yagi et al defined PJF as any form of symptomatic PJK that required surgery and reported a rate of only 1.4%.[9]

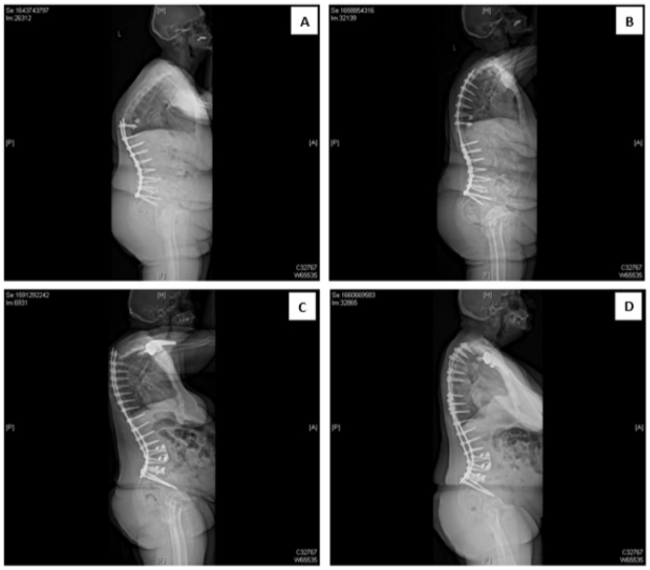

Figure 1. (A) Prerevision thoracolumbar proximal junctional failure (PJF). (B) Postrevision thoracolumbar PJF. (C) Prerevision proximal thoracic PJF. (D) Postrevision proximal thoracic PJF.

While the approach by Yagi et al helps to focus clinical research energies around PJFs most associated with increased EOCs, it runs the risk of excluding some cases with significant negative effects on PROMs who have not yet undergone surgical intervention. For example, while developing the PJF classifications proposed by Hart and Hostin from the ISSG database,[13,14] there were a significant number of PJF patients who were pending but had not yet undergone revision surgery that would have been excluded from the PJF analysis.

CLINICAL IMPACT OF PJK

Wide disparity in the reported clinical implications of PJK in the spinal deformity population is largely driven by the varied definition of the complication. Small incremental changes in adjacent segment kyphosis that use the definition of PJK based on accuracy of measurement of sagittal plane radiographs have largely found little adverse effect on PROs or reoperation rates.[3,5,7,10,16,17] Large multicenter pediatric deformity case series tend to be heavily weighted toward AIS. Hariharan et al reported a 6.0% all cause reoperation rate in 282 AIS patients at 10-year follow-up, and Dong et al, in an AIS series of 1816 patients, reported a 2.8% reoperation rate with an average follow-up of 8.5 years, with neither series reporting high rates of revision surgery for PJK.[18,19] In contrast, pediatric case series, which include the full spectrum of pediatric deformity, have reported 90-day reoperation rates as high as 14% but did not find PJK as a major driver of reoperation or readmission.[20] Findings such as these could lead to false conclusion that PJK is an incidental radiographic finding with little clinical significance.

Refuting the relegation of PJK to an incidental finding are studies that demonstrate decreased self-image scores in PJK patients,[21] lower SRS scores,[22] increased pain,[23] severe neurological injuries[9–11,24] as well as some series that reported PJK accounting for more than 50% of readmissions following treatment of ASD.[25]

PJK IMPACT ON EPISODE OF CARE COSTS

Evaluating the impact of PJK on spinal deformity episode of care costs (EOCs) is more straightforward than attempts to make more broad statements about its impact on purely clinical outcomes. By focusing research efforts on its contribution to readmission and reoperation, the varied definitions of PJK and PJF become less impactful. McCarthy et al, in their retrospective review of 184 ASD patients who underwent surgical correction between 2005 and 2011 with an average follow-up of 5 years, reported that the average total cost of hospitalizations for index operation to be \$103,143 ± \$39,655. The average total cost of care increased to \$120,394 ± \$60,820 due to costs associated with readmissions postoperatively, which averaged \$67,260 ± \$63,250. The readmissions accounted for a 74% increase in the total EOC and were largely driven by costs associated with revision surgery.[26] Authors from this study, however, did not list the indications that contributed toward the readmission and revision surgery costs. In an another study that included a total of 1218 consecutive ASD surgeries across 10 deformity centers, Hostin et al identified a PJF rate of 5.6% with failures occurring primarily in the thoracolumbar region of the spine. They found evidence that the mode of failure differs depending on the location of UIV, with thoracolumbar failures more likely due to fracture and upper thoracic failures more likely due to soft tissue failures. In a retrospective review of 695 consecutive ASD surgeries performed at a single center with a mean follow-up of 5.9 years, Yeramaneni et al reported an overall readmission rate of 24% with an average index surgical direct cost of \$86,081 and \$38,754 for readmissions. Of the 6 categories of readmission in their study, PJK readmission and reoperation occurred in 4.3% of the ASD cohort and was the second most common reason for readmission, accounting for 19.2% of the total readmissions, behind only infection at 24.4% in

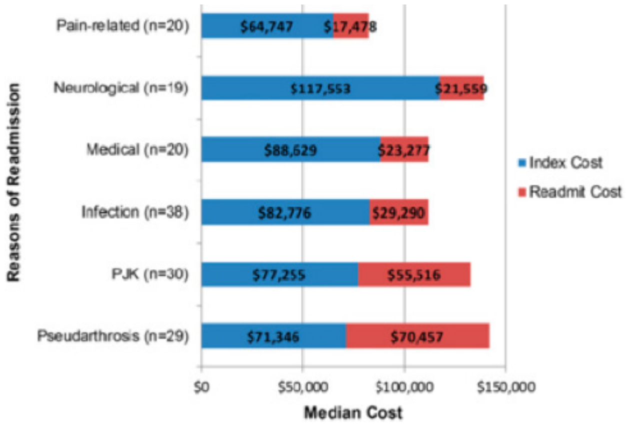

Figure 2. Direct costs associated with index surgery for adult spinal deformity and readmissions by indication. PJK, proximal junctional kyphosis. *Source:* Reprinted with permission form Yeramaneni et al.[27]

incidence. PJK was the second most expensive cause of readmission at $55,516 behind only pseudarthrosis at $70,457 (Figure 2). Their multivariate analysis demonstrated that each readmission for PJK was associated with a 63% higher readmission cost compared with readmissions due to medical reasons, and PJK resulted in roughly $2 million of additional EOCs for the cohort over the study period.[27] Their findings are consistent with costs reported from several additional studies. Hart et al, in a series of thoracolumbar fusions to the pelvis, reported an average inpatient cost of $77,432 for 2 patients who underwent reoperation with proximal extensions of their posterior fusions for PJK.[24] Safee et al reported direct costs of $119,217 ± $94,212 and total costs of $193,277 ± $152,613 in a cohort of 18 patients who underwent revision surgery for treatment of PJK.[28,29] Theologis et al reported an average direct cost of $55,547 ± $15,358 for revision operations for PJK over a 10-year period. They further found that revision operations for PJK in the lower thoracic spine had similar direct costs to revision operations for PJK in the upper thoracic region.[30]

Due to the high costs of revision surgery and its frequency as a driver of reoperation, optimizing the value of spinal deformity surgery will require reduction in the rates of symptomatic PJK. Further concern arises regarding the problematic nature of PJK, as supported by several studies indicating that despite the high cost of revision surgery, there is a high rate of recurrence of PJK (re-PJK) in patients with ASD after undergoing revision surgery for symptomatic PJK. Funao et al reported a 31% incidence of re-PJK and identified large initial PJA, high preoperative thoracic kyphosis and sagittal vertical axis (SVA), and greater correction of thoracic kyphosi and SVA as risk factors.[31] Kim et al[32] reported re-PJK rates of 44.3% in their cohort and found that prerevision thoracic pelvic angle and prerevision C2-T3 SVA were independent predictors of re-PJK.

PJK—Risk Factors and Prevention Strategies

The recognition of symptomatic PJK as a major driver of readmission, reoperation, and increased EOC has focused attention on strategies to reduce the occurrence of these junctional failures. These prophylactic strategies can be roughly grouped into 3 broad categories: biomechanical strategies, alignment/planning, and patient optimization/analytics. In a recent systematic review of the evidence for PJK prevention in ASD, Shlobin et al concluded that among the heterogeneous PJK prevention strategies reviewed, high-level evidence regarding any particular technique was limited.[33] In an analysis of the evolution of PJK and PJF rates over a 10-year period from the ISSG ASD database, Alshabab et al found that despite extensive research examining risk factors for PJK/PJF and increasing utilization of intraoperative PJK prophylaxis techniques, the rates of radiographic PJK and/or PJF did not significantly decrease across the 10-year enrollment period.[34]

Biomechanical Prophylactic Strategies

Using hooks instead of pedicle screw at the UIV to minimize the risk of PJK has been suggested extensively with conflicting results. The theoretical advantages of proximal hooks include a softer landing with lower mechanical stresses on the UIV as well as better maintenance of the vertebral body and less soft tissue dissection required for hook insertion. Several authors have reported decreased rates of PJK using hooks at the UIV,[5,22,35,36] while others have not found utilization of hooks at the UIV to decrease PJK rates.[17,37] From a health economic standpoint, UIV hooks have appeal due to their reduced cost relative to pedicle screws, but their role in the prevention of reoperation or positive effect on PRO is less than convincing. Additional attempts at decreasing construct stiffness by using a lower density of pedicle screws,[38] by reducing rates of anterior and posterior surgery,[16,21] and by using transition rods[39,40] or less stiff rods[41,42] have also been suggested, but much like utilization of hooks, their effects on reoperation rates and patient outcomes are not definitive.

Attempts to minimize posterior ligamentous failure by reinforcing the proximal interspinous ligament with Mersilene tape have also had mixed results, with some authors reporting PJK reductions using this technique[43–45] and another observing no difference in PJK rates with its use.[46] Pham et al and Alluri et al have explored ligament augmentation using tendon allograft with reduced rates of PJF in their small series.[47,48] Additional positive results using a variety of posterior polyester tethering techniques have also been reported.[49–53] Conclusions regarding the most appropriate indications for tether use, the optimal tether technique, as well as accumulation of enough data to do a cost-benefit analysis are still evolving.

Prophylactic vertebral cement augmentation has also been evaluated as a biomechanical means to reduce PJF and PJK in the ASD population. Hart et al, Martin, and Theologic et al have reported decreased PJK and PJF rates utilizing this technique.[24,54,55] A 5-year follow-up study by Raman et al found that prophylactic vertebroplasty, while minimizing the risk for junctional failure in the early postoperative period, did not appear to decrease the incidence of PJK at 5 years.[56] An additional study by Han et al found that while 2-level vertebroplasty did not decrease overall rates of PJK, it did appear to reduce rates of PJK progression and PJF.[57] In a cost-analysis study of prophylactic vertebral augmentation, Hart et al found the estimated cost to prevent a single proximal junctional acute collapse was $46,240 using vertebroplasty compared with inpatient costs of $77,432 associated with a revision instrumented fusion; thus, they suggested the technique may be cost-effective if utilized in elderly female patients undergoing extended lumbar fusions.[24] Further research is needed to examine which patients may receive the greatest benefit from cement augmentation based on preoperative risk factors as well as the long-term implications on degenerative changes that might be accelerated by the augmentation technique.

Alignment/Planning

Although an exhaustive review of the myriad of radiographic and alignment parameters that have been correlated with increased PJK and PJF incidence is beyond the scope of this review, it is clear that surgeries involving significant sagittal realignment are most at risk for developing clinically significant PJK and risk for reoperation in both pediatric and adult deformity reconstructions. It is also clear that there are not a few simple radiographic parameters that have proven consistently predictive of PJK. In the pediatric population, Scheuermann's kyphosis reconstruction cases account for a much higher rate of symptomatic PJK cases than AIS surgery, with Lowe and Kasten warning of the hazards of PJF with correction of kyphotic deformity by more than 50% nearly 30 years ago.[1]

In the adult population, long fusions to the pelvis for treatment of moderate to severe sagittal plane imbalance pose the greatest risk for junctional failures likely due to high biomechanical stresses from reciprocal change which is solely concentrated at the proximal end of the constructs. As in the pediatric population, overcorrection can significantly increase the risks of PJF and revision surgery. The recognition by Lafage et al that optimal alignment varies with age and the recognition of the importance of age-adjusted alignment goals with subsequent modification of the SRS-Schwab classification system is one of the most important recent spinal deformity scientific contributions.[58]

The European Spine Study Group developed Global Operation and Proportion (GAP) scores in order to help predict mechanical complications, including PJK, with conflicting studies on its utility in doing so.[59–61] GAP score parameters were relative pelvic version (the measured minus the ideal sacral slope), relative lumbar lordosis (the measured minus the ideal lumbar lordosis), lordosis distribution index (the L4–S1 lordosis divided by the L1–S1 lordosis multiplied by 100), relative spinopelvic alignment (the measured minus the ideal global tilt), and an age factor. ISSG more recently developed the Sagittal Age-Adjusted Score (SAAS). The score is composed of 3 sagittal parameters (PI–LL, PT, and and T1PA). For these 3 parameters, points are assigned based on offset from age-adjusted targets, and zero points were granted if the parameter was within a 10-year window above and below the patient's age. One point was added or subtracted for each 20-year window above or below the Match range. The total SAAS score was calculated by adding the score of each component (PI–LL, PT, and T1PA), with a negative score indicating under correction and a positive score indicating overcorrection. This system showed that higher SAAS scores were correlated with both higher PJK rates and more severe PJK.[62]

The importance of optimizing alignment is critical from a health economic standpoint because optimized planning adds little to no incremental cost to the index procedure and has the potential to significantly decrease what is one of the most expensive causes of readmission with its associated negative impact on cost per quality-adjusted life year (QALY). Passias et al showed failure to optimize sagittal alignment at the time of revision surgery for PJK led to worse clinical outcomes compared with patients who had their abnormal lumbopelvic mismatch corrected in combination with proximal extension of the fusion construct and showed increased rates of PJK and PJF in the malaligned group.[63] It is also unlikely that biomechanical prophylactic strategies designed to prevent PJK and PJF will be able to offset significant errors in alignment, rendering health care dollars utilized in their deployment futile.

PATIENT FACTORS/ANALYTICS

Many patient-specific risk factors for PJK have been identified, including low bone mineral density,[10,23,64] older age,[7,21,23,50,58] presence of a comorbidity,[7] male sex,[35] and sarcopenia.[64,65] In addition to the factors above, the type and severity of deformity as well as global measures of health such as frailty[66] clearly contribute strongly to PJK risk. The large number of patient-specific variables associated with risks of PJK will require employment of more sophisticated modeling and analytic tools and use of machine learning to aid in data integration and analysis. Scheer et al developed a preoperative model that is 86.3% accurate in predicting patients at risk of developing PJK using more than 60 patient-specific variables, with the 7 strongest predictors being age, lower-instrumented vertebra, preoperative SVA, UIV implant type, UIV, preoperative pelvic tilt, and preoperative lumbopelvic mismatch.[67] The development of increasingly accurate, easy to use, and widely available decision analytics tools will be critical to reducing the reoperation costs associated with symptomatic PJK and will require expansion of detailed quality multicenter data to drive their development.

CONCLUSIONS

Operative treatment of ASD results in improvements in multiple patient-reported outcomes, including SRS pain, SRS function, SRS self-image, and Oswestry Disability Index scores when comparing preoperative and 2-year postoperative states and in comparison to nonoperative management.[68] Glassman et al demonstrated that in patients with symptomatic ASD at 3-year follow-up, surgical patients experienced a gain of 2.3 QALYs for $111,451 compared with 0.4 QALYs for $29,124 with nonoperative care, with the incremental cost-effectiveness ratio of $44,033 in favor or surgical intervention.[69]

Despite these positive findings, pediatric spinal deformity and ASD surgery continues to struggle against both high index surgical costs and, particularly in the case of adult reconstruction, against high reoperation rates. In an increasingly value-based health care system, it is critical to endeavor to drive down index surgical costs, optimize patient outcomes, and increase the durability and long-term cost-effectiveness of these procedures. A great deal of recent literature has focused symptomatic PJK and PJF with frustratingly little reduction in the complication occurrence. To minimize the impact of junctional failures on the cost-effectiveness of pediatric and ASD surgery, research efforts should focus on patients who require reoperation within 2 years. Given widely variable patient and deformity types, development and testing of prevention strategies will require analysis of large, granular multicenter data sets and development of validated data analytic tools.

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Funding: The authors received no financial support for the research, authorship, and/or publication of this article.

Declaration of Conflicting Interests: J.G.: Consultant: Acuity, Depuy, Medtronic, Nuvasive, Stryker, FYR Medical; Royalties: Acuity, Medtronic, Nuvasive; Advisory Board: Medtronic, Stryker; Medical Board: National Spine Health Foundation; Stock: Cingulate Therapeutics, FYR Medical; Patent: Medtronic; Research: Stryker, Cerapedics, Inc., Biom'Up, empirical Spine, Pfizer, Texas Scottish Rite Hospital, Alan L. & Jacqueline B. Stuart Spine Research, Scoliosis Research Society, National Spine Health Foundation; Staff: Norton Health Care, Inc.; Honorarium: Baxter, Broadwater, NASS, Pacira Pharmaceuticals.

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