Published in final edited form as:

J Pediatr Orthop. 2019 April ; 39(4): 202–208. doi:10.1097/BPO.0000000000000898.

Distal Junctional Failure following Pediatric Spinal Fusion

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Abstract

Background—Adjacent segment pathology is a known complication after spinal fusion, but little has been reported on junctional failure. A series of adolescent patients presented with acute distal junctional failure. We sought to determine any common features of these patients in order to develop a prevention strategy.

Methods—A retrospective review was conducted of pediatric patients who developed distal junctional failure after instrumented spinal fusion performed at two institutions from 1999–2013. Patients with proximal junctional failure or junctional kyphosis without failure were excluded.

Results—Fifteen subjects were identified with mean follow-up of 38 months. Distal failure occurred a mean of 60 days after index surgery, with history of minor trauma in four patients. Failures included 3-column Chance fracture (11) or instrumentation failure (4). Thirteen patients presented with back pain and/or acute kyphosis, while two asymptomatic patients presented with healed fractures. Two patients also developed new onset of severe lower extremity neurologic deficit after fracture, which improved but never resolved after revision. 13/15 subjects required revision surgery, typically within one week. Complications associated with revision surgery were encountered in 8 patients (62%). Major complications that required return to the operating room included two deep infections, two instrumentation failures, and dense lower extremity paralysis that improved after medial screw revision and decompression. At final follow-up, 10 patients are asymptomatic, two have persistent neurologic deficit, two have chronic pain, and one has altered gait with gait aid requirement.

Conclusion—This study analyzes a heterogeneous cohort of spinal fusion patients who developed distal junctional failure from 3-column Chance fracture or instrumentation failure. Revision surgery is typically required, but has a high complication rate and can result in severe neurologic deficit, highlighting the morbidity of this complication. It is unclear whether level of the lowest instrumented vertebra contributes to distal junctional failure. Increased awareness of

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Level of evidence: IV Study Type Case series

CONFLICT OF INTEREST: Dr. Rathjen reports royalties from Elsevier, stock/stock options from Mati Therapeutics. Dr. Larson's institution receives/has received research support from Scoliosis Research Society and Orthopedic Research and Education Foundation.

junctional failure in children may prompt additional studies to further characterize risk factors and preventative strategies.

INTRODUCTION

Posterior instrumented spinal fusion is an established treatment for pediatric patients with spinal deformity. Although this is generally considered a safe procedure, serious short-term complications following pediatric spinal fusion are unfortunately common [1–4]. Reames et al reviewed 19,360 pediatric patients in the Scoliosis Research Society (SRS) database and found an overall complication rate of 10.2% for the surgical treatment of scoliosis [5]. Likewise, Coe et al reviewed the SRS database, and found an 11.8% complication rate amongst 499 pediatric patients treated with spinal fusion for Scheuermann's kyphosis [6]. A thorough understanding of complications is imperative for preoperative counseling, surgical decision-making, and post-operative care.

Adjacent segment pathology is a known complication that can occur after spinal fusion surgery. Junctional deformity can be acute or chronic, and oftentimes is asymptomatic and can be observed unless failure occurs. Late deformity can often be related to curve progression adjacent to the fused segment [7]. Adjacent segment pathology is a broad term that encompasses a spectrum of disease, including junctional scoliosis (JS), proximal junctional kyphosis (PJK), distal junctional kyphosis (DJK), proximal junctional failure (PJF), and distal junctional failure (DJF).

Risk factors for adjacent segment pathology include improper selection of instrumentation levels and disruption of anatomic structures [7–12]. Additionally, patients with pre-existing sagittal plane deformity may be at higher risk for junction problems [11–14], and overcorrection or undercorrection of sagittal deformity can also contribute to junctional disorders [9, 15, 16]. Frequently, the etiology of junctional deformity is not well-understood.

Distal junctional kyphosis has been associated with choice of lowest instrumented vertebra (LIV) after fusion for thoracic kyphosis [9, 14, 15, 17–20]. Previous authors have advocated for choosing a LIV to include the first lordotic vertebra (FLV), defined as the vertebra immediately caudal to the first lordotic lumbar disc [15, 17, 21]. Other authors have proposed that fusion should be extended to the stable sagittal vertebra (SSV). Cho et al defined the SSV as the most proximal vertebra touched by the posterior sacral vertical line (PSVL), which is a line drawn vertically from the posterior corner of the superior sacral endplate on a lateral upright radiograph [9, 14]. Fusion to the SSV was found to be superior at preventing DJK when compared to fusion to the FLV [9, 14, 19].

The LIV in scoliosis has traditionally been chosen based on coronal radiographs, with fusion to the end vertebra, stable vertebra, or last vertebra touched by the CSVL based upon the individual curve characteristics [20]. However, the role of sagittal balance and its association with junctional deformity is gaining more attention. Broom et al recently presented data analyzing DJK in scoliosis patients. This group used a slightly different definition of the SSV, as they described it as the most proximal vertebra with 50% or more of its body

anterior to the posterior sacral vertical line. This group found that inclusion of the SSV prevented DJK in fusion of patients with AIS [22].

Arlet and Aebi reviewed junctional spinal disorders in adults, and described the most common distal failure modes as shown in Table 1 [7], but there is minimal description in the literature of junctional failure in the pediatric population.

We have noted a series of adolescent patients who presented with 3-column “Chance” fracture or instrumentation failure at the distal end of a spinal fusion construct within the first three months following index surgery. Although this mode of failure has been reported in the adult population [23, 24], it is rare in the pediatric literature. The purpose of our multicenter study was to retrospectively review the features of distal junctional failure in pediatric patients following instrumented spinal fusion. We sought to determine any common features of these fractures in order to develop a strategy to prevent this complication.

MATERIALS/METHODS

After Institutional Review Board approval, a retrospective review was conducted of pediatric and adolescent patients who had undergone instrumented spinal fusion at two institutions. Among 1700 pediatric spinal fusion cases performed from 1999 – 2013, we identified 15 patients (15/1700; 0.9%) who developed acute distal junctional failure. Patients with proximal junctional kyphosis and failure were excluded, as were patients with distal junctional kyphosis without failure. The minimum clinical and radiographic follow-up period was set at 12 months. All underlying spinal disorders prompting elective spinal fusion (including idiopathic and neuromuscular scoliosis, kyphosis, and spinal stenosis) were included in this analysis. Two reviewers (LF, AS) who were not involved in the surgical treatment performed standardized data abstraction.

Demographic information was collected, including age at index surgery, gender, body mass index (BMI), and underlying diagnosis. Preoperative radiographs were analyzed for major curves, curve classification, and sagittal balance. Procedures were carefully reviewed, and data collected included: upper and lower instrumented levels, integrity of the distal interspinous ligament, whether or not the pedicles were tapped, neuromonitoring status/changes, decortication, instrumentation system, and other procedures performed at index surgery in addition to posterior spinal fusion. After index surgery, the date of failure, method of diagnosis, treatment, and complications were carefully analyzed. Final clinical and radiographic outcomes were reviewed.

RESULTS

15 pediatric spinal fusion patients, including 10 females and 5 males, were included in this study. Mean follow up was 38 months (range 13–95 months). The mean BMI was 25 kg/m² (14 – 42), and the mean age at index surgery was 14 years (7–19). Six surgeons performed the fusions at two separate institutions from 1999 to 2013. The patient demographics and diagnoses for all subjects are shown in Table 2.

All preoperative, postoperative, and post-revision radiographs were closely examined (Table 3). For the 10 scoliosis patients, fusion was extended to the stable vertebra in 2 patients and to the lowest vertebra touched by the center sacral vertical line (CSVL) in 3 patients, while 4/10 patients were fused cephalad to both of these levels. 5/10 patients were fused to the SSV as described by Broom [22]. Amongst the 6 patients with kyphotic deformity, 4 were fused to the SSV as described by Cho et al [9], while 2 were fused one level caudal to the SSV.

In all patients, the distal interspinous ligament was left intact. Seven different types of instrumentation systems were used, including use of titanium (7), stainless steel (5), and cobalt chrome (3). Hook-only constructs were used in two patients, whereas pedicle screws were used in 13 cases, with hooks at the most cephalad level in 10 of 13 patients. The LIV was decorticated in all cases. At one of the institutions, the pedicles were not tapped prior to screw insertion in 8 of 9 cases.

There were no intraoperative complications during index spinal fusion. One patient had preoperative neurological deficits with no lower extremity somatosensory evoked potentials, so neuromonitoring was not performed. In the remaining patients, there were no intraoperative neuromonitoring changes other than minor arm changes attributed to patient positioning. No neurologic changes were noted in the initial postoperative phase.

Diagnosis of distal junctional failure was noted on average 60 days after index surgery (range 3–119). There was a history of fall or injury in 4 patients, all of which were relatively minor accidents. 7/15 presented within 30 days of index surgery, while 8/15 presented after 30 days. Two patients were asymptomatic and presented at routine follow-up three months after index surgery with distal bony Chance fractures that had already healed in kyphosis. The remaining 11 patients presented with back pain and/or acute kyphosis.

After Chance fracture, two patients developed new neurologic deficits. A patient with Scheuermann's kyphosis was noted to have incomplete neurologic loss on postoperative day 3, after L3 Chance fracture. The second patient with neurologic changes had stenosis due to achondroplasia, with preoperative weakness distal to her knees bilaterally and urinary incontinence. Her neurologic examination was initially unchanged after index surgery, but 29 days postoperatively, her parents reported new onset of bowel incontinence and worsened left lower extremity weakness. Radiographs revealed L2 chance fracture with kyphotic deformity that compromised the spinal canal.

Of the 15 distal junctional failures, 11 (73%) were 3-column bony Chance fractures (Fig. 1, Fig. 2), while four were related to instrumentation failure with implant loosening or disengagement (Fig. 3). All of the 3-column Chance fractures occurred through the LIV or at the junction of the LIV and adjacent segment. A mean of 30 degrees focal kyphosis occurred between the LIV and distal adjacent segment, which prompted CT scan to further visualize the pathology and for preoperative planning for revision surgery.

Thirteen of 15 patients underwent revision surgery to address the distal junctional failure. Two patients who presented with healed Chance fractures were managed nonoperatively. 11 revision surgeries were performed within one week of diagnosis, one performed at two

weeks, and one performed at four months after a failed trial of extension casting. The pathologic kyphosis was reduced in all revision cases, with additional procedures as noted in Table 2. Two patients had noted improvement in SSEP/MEP potentials by the end of the revision case, with no worsening signals intraoperatively.

Of the two patients with neurologic decline after chance fracture, both had noted improvement after revision surgery, but neither fully recovered. The Scheuermann's kyphosis patient was noted to have a grossly unstable fracture with a large fracture hematoma, and had significant improvement in SSEP/MEP potentials by the end of the revision case. Postoperatively, he regained full strength and sensation, but had chronic right-sided L3 radicular pain. The achondroplasia patient had improvement in strength after revision, but did not regain bowel function.

As a result of revision surgery, complications were encountered in 8/13 patients (62%). Minor complications that did not require additional surgery included intraoperative dural tear, and superficial wound dehiscence that healed uneventfully with conservative management. One patient developed a delayed pleural effusion that required chest tube placement. Major complications requiring return to the operating room were encountered in 5 patients (38%), including 2 patients with implant failure, 2 with infection, and 1 patient with neurologic decline (see Table 2). This patient developed dense lower extremity paraparesis in the days following revision surgery, prompting return to the operating room for decompression. The left L4 screw was noted to be medial, and was revised accordingly. At follow-up, this patient had regained the ability to ambulate with a walker but had residual neurogenic bowel and bladder.

Final clinical follow-up occurred at a mean of 38 months (range 13–95 months). At follow-up, 10 patients (67%) were asymptomatic. Two patients (13%) have residual neurologic injuries, as described above; although they regained ability to ambulate with a walker, both have residual neurogenic bowel and bladder. Two patients (13%) have chronic pain, one with paraspinal pain (mild) and one with chronic L3 radicular-type pain (mild). One patient (7%), who was treated nonoperatively for a healed Chance fracture, now ambulates with a crouched gait due to forward stance from kyphosis, and requires a wheelchair for long distances.

DISCUSSION

Distal junctional failure from 3-column Chance fracture or instrumentation failure is a rare but devastating complication following spinal fusion surgery. 88% of our cohort required revision surgery for deformity reduction, instrumentation revision with fusion extension, osteotomy and/or neural decompression. Three patients (20%) had resultant neurologic deficits; two after Chance fracture through the LIV, and one resulting from revision surgery. While all three patients improved after revision, two have residual neurologic deficits while the third has chronic radicular pain. Deep surgical site infection, pleural effusion requiring chest tube, and repeat instrumentation failure were also major complications encountered in our small cohort. Clearly this complication has significant consequences and additional studies are needed to help clarify patient and surgical risk factors and preventative strategies.

Distal junctional failure has not been well-studied in the pediatric or adolescent literature. In a metaanalysis of complications in neuromuscular scoliosis patients, Sharma et al. found the rate of implant breakage to be 4.6%, while the rate of screw cutout, pullout, or migration was 2.4% [4]. Additionally, large series investigating all complications resulting from spinal fusion in pediatric and adolescent patients have identified acute junctional failure resulting from implant failure, oftentimes in the neuromuscular population [4, 25, 26]. However, detailed analysis of distal junctional failure has not been well described.

While there are reports in the pediatric and adolescent population on proximal junctional kyphosis [12, 13, 27] and distal junctional kyphosis [16, 28], junctional failure studies are better studied in the adult literature. Proximal junctional failure in adults has been associated with osteopenia, preoperative sagittal imbalance, marked correction of sagittal malalignment, and patient comorbidities [8, 10, 11, 29–31].

Distal junctional failure studies are less common. In 1998, Levine et al. reported a 44 year old male with neuromuscular kyphosis who sustained a Chance fracture at the caudal end of a pedicle screw fixation construct. This patient required fracture reduction and extension of the fusion to the pelvis to restore sagittal balance [23]. Kwon et al. subsequently evaluated sagittal decompensation and distal junctional failure in 13 adults. Patient risk factors included osteoporosis, hip osteoarthritis, and substance abuse, all of which likely are less relevant in pediatric patients. Revision in this series was typically achieved using pedicle subtraction osteotomy and fusion extension to the sacrum [23].

Distal junctional kyphosis has been associated with improper selection of vertebral levels. The choice of LIV for fusion of Scheuermann's kyphosis or other thoracic hyperkyphosis is typically based on sagittal radiographs [9, 14, 15, 17–19, 21]. Previous authors have advocated for choosing a LIV to include not only the end vertebra of the kyphosis, but also the first lordotic vertebra (FLV) [15, 17, 21] or the stable sagittal vertebra (SSV) [9, 14]. Fusion to the SSV has been found to be superior at preventing DJK when compared to fusion to the FLV [9, 14, 19].

Conversely, the LIV in scoliosis has traditionally been chosen based on coronal radiographs, with fusion to either the end vertebra, the stable vertebra, or the last vertebra touched by the CSVL based upon the individual curve characteristics [20]. However, the role of sagittal balance and its association with junctional deformity has recently drawn more attention. Broom et al presented data analyzing DJK in scoliosis patients. This group described the SSV as the most proximal vertebra with 50% or more of its body anterior to the posterior sacral vertical line, and found that inclusion of this level prevented DJK in fusion of patients with AIS [22].

It is unclear whether distal junctional failure is associated with choice of LIV. In our radiographic review, we found that all patients with preoperative kyphotic deformity were fused to an appropriate level, in that they were fused at or caudal to the SSV described by Cho et al [9]. This includes patients with both scoliosis and hyperkyphosis (subjects 9 and 10 in Table 3). Thus, we cannot attribute distal junctional failure to the choice of LIV in our cohort of kyphosis patients.

Conversely, we found that 4/10 (40%) of the scoliosis failures were fused cephalad to both the stable vertebra and to the lowest vertebra touched by the CSVL in the frontal plane. Additionally, 5/10 (50%) were fused cephalad to the SSV as described by Broom [22]. Only 3/10 (30%) were fused to appropriate levels on both coronal and sagittal radiographs, namely the last level touched by the CSVL and the SSV described by Broom. Despite appropriate choice of LIV by both frontal and sagittal radiographic parameters, these scoliosis patients still developed distal junctional failure, thereby indicating that other factors are involved.

There are several limitations to this study, the first of which is its retrospective nature. Additionally, our cohort was drawn from two centers, and includes many surgeons who utilized several different techniques, making it difficult to draw definitive conclusions as to the risk factors and preventative strategies associated with this complication. Another limitation of our study is the small number of subjects, despite the broad inclusion criteria from two centers; this demonstrates the rarity of this complication. Although we initially attempted a matched cohort for further analysis, we were unable to do so effectively, given the heterogeneous nature of our subjects.

As a result of this study showing that 8/9 subjects at one institution occurred in patients in whom pedicles were not tapped prior to screw placement, the surgeons who previously did not tap pedicles prior to screw placement now do so. Tapping may help dilate the pedicle to avoid inadvertent stresses and occult fracture of the pedicle during screw placement, which could later result in screw pullout or Chance fracture. Additionally, we have discontinued aggressive decortication at the LIV as a precaution to avoid structurally weakening the LIV. We also now give consideration to postoperative bracing for patients with large pre-operative kyphosis, or with a large amount of sagittal deformity correction during fusion.

This study of distal junctional failure in the pediatric population may draw attention to this major complication following spinal fusion. More awareness of junctional failure in children may prompt additional studies to further characterize risk factors and preventative strategies.

Acknowledgments

FUNDING: ANL was supported by an NIH from the National Institute of Arthritis and Musculoskeletal and Skin Diseases. (R03 AR 66342). No other support was received for this manuscript.

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Figure 1.

11 year old female with AIS who sustained 3-column chance fracture through the inferior endplate of L1, occurring 76 days after T2-L1 fusion. She underwent L1–2 laminectomy and discectomy, transforaminal lumbar interbody fusion, fracture reduction, and L1–L4 fusion extension.

Figure 2.
12 year old female with juvenile idiopathic scoliosis who was diagnosed with a three-column fracture one month following posterior spinal instrumentation. Due to a chronic pars fracture at L5, she underwent extension of her fusion to the pelvis.

Figure 3.

16 year old female with Scheuermann's kyphosis who fell one month after T3 to L3 fusion and developed distal instrumentation failure with right pedicle screw pull-out and left rod disengagement. She underwent L2 foramen decompression and revision posterior fusion with extension to L4.

Table 1

Modes of distal junctional failure, as described by Arlet and Aebi [7]

Modes of Distal Junctional Failure
1. Progressive loss of lumbar lordosis, disc degeneration with loss of height
2. Acute wedging in disc below instrumentation
3. Fracture of distal instrumented vertebra
4. Osteoporotic fracture below long rigid fixation
5. Failure of instrumentation at most distal level
6. Spinal stenosis and/or segmental instability below instrumentation

Table 2

Patient demographics and clinical course.

	Age	Sex	BMI	Diagnosis	Pathology	Year of Index Surgery	Procedure Performed	Pathology	Revision	Additional Revisions
1 *	11	F	28	AIS	Scoliosis	2011	PSF T2-L1	L1–2 Chance fracture	Revised instrumentation L1–L4, decompression & TLIF L1–L2	
2	16	F	29	AIS	Scoliosis	1999	PSF T2-L2	L2 Chance fracture	Revised instrumentation L1–L2	Distal screw pullout, PSF revision with extension to L3
3	18	M	14	AIS	Scoliosis	2013	PSF T3-L3	Distal implant disengagement	Revised instrumentation L1–L4	–
4	11	F	27	AIS	Scoliosis	2012	PSF T2-L1	L1 Chance fracture (healed)	–	–
5	13	F	27	AIS	Scoliosis	2005	PSF T4–T13, thoracoplasty	Acute distal implant loosening	Revised instrumentation, extension to L1	–
6	11	F	24	AIS	Scoliosis	2007	PSF T5–L1	L1–2 pedicle fracture	Revised instrumentation, extension to L3	
7	15	M	25	William’s Syndrome	Scoliosis	2003	PSF T2-L1	L1 Chance fracture	Revised instrumentation T12-L2	
8	12	F	20	Chromosome deletion 1p36	Scoliosis	2012	PSF T2-L1	L1 Chance fracture (healed)	–	–
9 **	12	F	16	JIS	Scoliosis/Kyphosis	2013	PSF T2-L1	L4 Chance fracture	Revised instrumentation to pelvis, L4 laminectomy	
10	14	M	15	Marfan’s Syndrome	Scoliosis/Kyphosis	2012	PSF T3-L4	Distal implant failure	Revised instrumentation, SPO T11-L4, extension to L4	
11	17	F	27	Marfan’s Syndrome	Kyphosis	2013	PSF T4-L5	L4 Chance fracture	Revised instrumentation, extension to L6	3 I&Ds for infection
12 ***	16	F	42	Scheuermann’s kyphosis	Kyphosis	2013	PSF T2-L4	Implant disengagement, L3 Chance fracture	Revised instrumentation L2–L4, foraminotomy L2	I&D for infection
13	15	M	38	Scheuerman’s kyphosis	Kyphosis	2009	PSF T3-L3	L3 Chance fracture	Revised instrumentation, hematoma decompression, extension to pelvis	2 subsequent implant revisions, I&D for seroma, screw revision for radiculopathy
14	19	M	25	Rubenstein Taybi	Kyphosis	2006	PSF T2-L2	L2 Chance fracture	Revised instrumentation L1-L4, SPO & discectomy L1-L2	Neurologic decline: revised PSF L1–L4, decompression L1–L2, foraminotomies L1–L3
15	7	F	21	Achondroplasia	Stenosis	2005	Decompression, PSF & ASF T10-L2, laminoplasty L2-S1	L2 Chance fracture	T10-L4 PSF revision, L1–L4 decompression, L2–L3 discectomy, transtheal extension osteotomy	–

(JIS – Juvenile Idiopathic Scoliosis, AIS – Adolescent Idiopathic Scoliosis, PSF – posterior spinal fusion, TLIF – transforaminal lumbar interbody fusion, SPO – Smith-Peterson osteotomy, I&D – irrigation and debridement).

* see Figure 1.

** see Figure 2.

*** see Figure 3.

Table 3

Radiographic findings

Patient	Lenke classification	PT curve	MT curve	TL curve	L curve	Kyphosis	Stable	Last touched by CSVL	SSV 1	SSV 2	LIV	Revision LIV	Scoliosis correction	Kyphosis correction
1*	2BN	43	72	0	56	28	L1	T12	L2	L1	L1	L4	72%	–
2	1AN	28	53	0	36	35	L3	L3	L4	L3	L2	L2	75%	–
3	5CN	17	33	0	53	32	L5	L4	L3	L2	L3	L4	83%	–
4	2AN	40	50	25	0	30	T13	T12	L2	S13	L1	–	86%	–
5	1BN	14	54	0	48	33	L2	L1	L1	T13	T13	L1	17%	–
6	1B+	30	50	0	30	44	L2	L1	L1	T12	L1	L3	80%	–
7	–	22	52	40	10	35	L1	T10	L2	L1	L1	L2	35%	–
8	–	29	70	0	23	22	L4	L3	T12	T11	L1	–	91%	–
9**	–	0	51	69	27	73	L5	L4	L4	L3	L4	Pelvis	64%	70%
10	–	0	52	67	13	54	L5	L4	L4	L3	L4	L4	58%	30% (+16° DIJ)
11	–	0	0	0	0	88	–	–	L5	L4	L4	L6	–	56% (+32° PIJ)
12***	–	0	0	0	0	95	–	–	L4	L3	L3	L4	–	23%
13	–	0	0	0	0	76	–	–	L4	L3	L3	Pelvis	–	39%
14	–	0	0	0	0	110	–	–	L3	L2	L2	L4	–	35%
15	–	0	0	0	0	48	–	–	L4	L3	L2	L2	–	40%
MEAN		22	54	20	30	54							66%	42%

PT – proximal thoracic (cephalad to MT), MT – main thoracic (T2-T11/12 disc), TL – thoracolumbar (T12-L1), L – lumbar (L1/2 disc-L4), CSVL – center sacral vertical line, SSV 1 – stable sagittal vertebra described by Broom et al (22), SSV 2 stable sagittal vertebra described by Cho et al [9], FLV – first lordotic vertebra, LIV – lowest instrumented vertebra

* see Figure 1.

** see Figure 2.

*** see Figure 3.