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Complications and Revisions After Spine Surgery in Patients With Skeletal Dysplasia: Have We Improved?

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Abstract

Study Design: Retrospective case series.

Objective: To report contemporary rates of complications and subsequent surgery after spinal surgery in patients with skeletal dysplasia.

Methods: A case series of 25 consecutive patients who underwent spinal surgery between 2007 and 2017 were identified from a single institution's skeletal dysplasia registry. Patient demographics, medical history, surgical indication, complications, and subsequent surgeries (revisions, extension to adjacent levels, or for pathology at a non-contiguous level) were collected. Charlson comorbidity indices were calculated as a composite measure of overall health.

Results: Achondroplasia was the most common skeletal dysplasia (76%) followed by spondyloepiphyseal dysplasia (20%); 1 patient had diastrophic dysplasia (4%). Average patient age was 53.2 $\pm$ 14.7 years and most patients were in excellent cardiovascular health (88% Charlson Comorbidity Index 0-4). Mean follow up after the index procedure was 57.4 $\pm$ 39.2 months (range). Indications for surgery were mostly for neurologic symptoms. The most commonly performed surgery was a multilevel thoracolumbar decompression without fusion (57%). Complications included durotomy (36%), neurologic complication (12%), and infection requiring irrigation and debridement (8%). Nine patients (36%) underwent a subsequent surgery. Three patients (12%) underwent a procedure at a non-contiguous anatomic zone, 3 (12%) underwent a revision of the previous surgery, and another 3 (12%) required extension of their previous decompression or fusion.

Conclusions: Surgical complication rates remain high after spine surgery in patients with skeletal dysplasia, likely attributable to inherent characteristics of the disease. Patients should be counseled on their risk for complication and subsequent surgery.

Keywords

degenerative, stenosis, myelopathy, deformity

Introduction

Osseous spine abnormalities are a common feature of many skeletal dysplasias, which are a heterogeneous group of >400 distinct genetic disorders affecting growth and development of the skeleton.[1] Achondroplasia is characterized by short, thickened pedicles; diastrophic dwarfism is associated with high rates of scoliosis and kyphosis, and spondyloepiphyseal dysplasia often exhibits cervical spine instability and congenital stenosis.[2-6] Such abnormalities predispose patients to compression of neural elements, leading to myelopathy or radiculopathy at a relatively early age. Concomitant spinal deformity may exacerbate the pathology or complicate the treatment algorithm. Intractable neurologic symptoms may mandate surgical intervention.

While these patients may experience progressive symptoms, surgical decision-making is a highly complex process, as spine

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procedures in patients with skeletal dysplasia have been associated with high rates of complications.[5-9] Atypical collagen leads to friable connective tissues, predisposing patients to dural tears and wound healing complications,[10] while unfamiliar anatomy may complicate the surgical approach or instrumentation, increasing the risk for neurologic injury. Furthermore, associated medical conditions place patients at high risk for cardiac or pulmonary complications, with prior case series demonstrating complication rates as high as 61% after spine surgery in patients with achondroplasia.[9] However, much of our current knowledge regarding spine surgery in this patient population is based on registries from only a few institutions, with some containing cases from almost 50 years ago.[2,4,9]

Contemporary, enhanced preoperative medical optimization and multidisciplinary perioperative care protocols have led to improved outcomes in medically-complex patient populations.[11-13] Expert opinion recommends such practices in the perioperative management of patients with skeletal dysplasias.[14] Care for patients with skeletal dysplasias at our institution is coordinated through a dedicated, multidisciplinary center. Collaboration between the treating spine surgeon and medical staff facilitates communication among care teams, optimizes safety, increases access for patients, and allows tracking of long-term outcomes. Preoperative evaluation and optimization are catered to both the patient's specific skeletal dysplasia diagnosis and individual medical needs. This includes pulmonary, cardiac, neurologic, and sleep testing, as well as case management and social work meetings prior to the planned surgery date. Rehabilitative care is managed by therapists and medical physicians familiar with conditions and complications associated with the skeletal dysplasia diagnosis.[15]

Given these advances in medical optimization, there is need to analyze contemporary rates of complications in this patient population. Furthermore, no series has reported rates of revision or need for subsequent spine surgery, which are relevant concerns for operative planning and patient counseling, given the frequency of pathology at multiple contiguous or non-contiguous levels. As such, the purpose of this study is to report rates of complications, revisions, or subsequent spine surgery in a contemporary series of patients with skeletal dysplasia. We hypothesized that complication rates after spine surgery would be lower compared to historical case series.

Materials and Methods

Institutional Review Board approval was obtained to study skeletally mature patients with a skeletal dysplasia who had undergone spine surgery between 2007 and 2017 at either 1 of 2 hospitals affiliated with a single institution (Hospital for Special Surgery, HSS). Participants were identified from a prospectively-enrolled skeletal dysplasia database maintained by the Center for Skeletal Dysplasias at HSS. Our institution is an orthopaedic hospital uniquely focused on treating patients with musculoskeletal conditions, including skeletal dysplasias. A detailed description of our skeletal dysplasia center's multidisciplinary process has been previously published.[15]

Out of the 39 patients identified from the database, 7 were excluded due to inadequate documentation of the operative procedure and postoperative course. Four patients who underwent prior spine surgery at outside hospitals were excluded. Two additional patients with multiple hereditary exostoses who underwent simple exostosis excision were excluded. One patient with a concomitant diagnosis of juvenile rheumatoid arthritis (JRA) was excluded. For the remaining 25 patients mean age was 53.2 $\pm$ 14.7 years and average body mass index (BMI) was 34.8 $\pm$ 8.9 kg/m^2 at the time of the index operation. Charleston comorbidity indices (CCI) were calculated for each patient as a composite measure of overall health and cardiovascular risk.[16-18]

The final case series included 19 (76%) patients with achondroplasia, 5 (20%) patients with spondyloepiphyseal dysplasia, and 1 patient (4%) with diastrophic dysplasia (Table 1). The most common indication for surgery was neurogenic claudication (n = 8 patients; 32%) followed by lumbar radiculopathy (n = 5; 20%), and cervical or thoracic myelopathy (n = 4; 16%). Seven patients reported preoperative bowel or bladder incontinence.

Mean follow up after the index procedure was 32.8 $\pm$ 42.4 months (range 2-169 months) Primary outcomes of interest were subsequent spine procedures (either revisions for failure of the index surgery, extension of the previous decompression or fusion, or for pathology at a non-contiguous level) and complications. Complications were grouped into the following categories as have been previously utilized in the literature: neurologic, thromboembolic, pulmonary, cardiovascular, gastrointestinal, genitourinary, intraoperative dural tears, infections at the operative site requiring irrigation and debridement, and mortality.[9] Neurologic complications were defined as any new postoperative deficit that did not resolve at final follow up.

Results

The patient population was relatively healthy at the time of surgery, with a CCI of 0-4 in 88% of the sample. Sixteen complications occurred in 12 patients (48%) (Table 2). Durotomy was the most common complication (n = 9; 36%), followed neurologic complication (n = 3; 12%) and acute infection requiring irrigation and debridement (n = 2; 8%). Neurologic complications included new onset right foot drop and left quadriceps weakness after a T12-S1 decompression (achondroplasia), bilateral quadriceps weakness after an L1-S1 decompression (achondroplasia), and worsening proximal upper and lower extremity weakness after a C1-3 decompression (spondyloepiphyseal dysplasia). There were 2 medical complications, both occurring in the same patient (Figure 1), who experienced a pulmonary embolus (PE) and a urinary tract infection (UTI) postoperatively.

Table 1. Cases Included in Series.

Patient	Age	Skeletal Dysplasia	Surgical Diagnosis	Indication	Surgical Procedure	Complication
1	60	Achondroplasia	Lumbar spinal stenosis	Neurogenic claudication	PD L1-L5	Durotomy
2	52	Achondroplasia	Lumbar spinal stenosis	Bladder incontinence	PD T12-S1	New neurologic deficit
3	52	Achondroplasia	Lumbar stenosis	Neurogenic claudication, bladder incontinence	PD T12-S1	
4	64	Achondroplasia	Lumbar spinal stenosis	Neurogenic claudication, bowel/bladder incontinence	PD L1-S1	
5	37	Achondroplasia	Lumbar spinal stenosis	Bilateral lumbar radiculopathy	PDF T10-L4	
6	75	Achondroplasia	Lumbar spinal stenosis	Neurogenic claudication	PD L1-S1	Durotomy, new neurologic deficit
7	15	Achondroplasia	Lumbar spinal stenosis	Cauda equina syndrome	PD T10-L3	
8	65	Spondyloepiphyseal Dysplasia	Herniated lumbar disc	Lumbar radiculopathy	PD L1-L2	
9	42	Achondroplasia	Lumbar spinal stenosis	Neurogenic claudication	PD L1-S1	
10	51	Achondroplasia	Lumbar spinal stenosis	Lumbar radiculopathy	PD L1-L5	Durotomy
11	59	Diastrophic Dysplasia	Lumbar spinal stenosis	Bilateral lumbar radiculopathy with neuro deficit	PD T12-S1	Durotomy, prolonged intubation
12	62	Achondroplasia	Lumbar spinal stenosis	Cauda equina syndrome	PD T12-S1	Durotomy
13	72	Spondyloepiphyseal Dysplasia	Cervical DDD	Cervical radiculopathy	ACDF C4-6	
14	34	Achondroplasia	Lumbar spinal stenosis	Neurogenic claudication	PD T12-L5	Durotomy
15	40	Spondyloepiphyseal Dysplasia	Lumbar spinal stenosis, spondylosis	Neurogenic claudication	PD T12-L3, PF T10-L3	
16	64	Spondyloepiphyseal Dysplasia	Os odontoideum	Cervical myelopathy	PDF C1-C3, suboccipital decompression	UTI, new neurologic deficit, pulmonary embolus
17	59	Achondroplasia	Lumbar spinal stenosis	Lumbar radiculopathy	PD L2-S1, PF L3-L4	Durotomy
18	67	Achondroplasia	Cervical Stenosis	Cervical myelopathy, lumbar radiculopathy	PD T8-T9, L5-S1	Infection requiring I&D
19	49	Achondroplasia	Thoracolumbar stenosis	Neurogenic claudication	PD T12-S1	Durotomy
20	44	Achondroplasia	Thoracolumbar stenosis	Neurogenic claudication	PD T12-S1	Durotomy, infection requiring I&D
21	10	Spondyloepiphyseal Dysplasia	Thoracolumbar scoliosis	Progressive deformity	PF T2-L3	
22	45	Achondroplasia	Thoracolumbar stenosis	Bowel/bladder incontinence, lumbar radiculopathy	PD T11-S1	
23	13	Achondroplasia	Lumbar spinal stenosis	Neurogenic claudication	PD L1-L3	
24	50	Achondroplasia	Lumbar spinal stenosis	Lumbar radiculopathy	PD T11-S1	
25	29	Achondroplasia	Lumbar spinal stenosis	Lumbar radiculopathy	PD L1-S1	

PD, posterior decompression; PDF; posterior decompression & fusion; PF, posterior fusion; ACDF, anterior cervical discectomy and fusion; ROH, removal of hardware; I&D, irrigation and debridement; UTI, urinary tract infection.

Nine patients (36%; 8 patients with achondroplasia, one with diastrophic dysplasia) underwent a second spine procedure during the follow up period (Table 3). The average time between the first and second surgery was 10.4 $\pm$ 11.8 months (range 0.4-39.4 months). Three patients (12%) underwent a procedure at a non-contiguous anatomic zone, 3 (12%) underwent a revision of the previous surgery, and another 3 (12%) required extension of their previous decompression or fusion (Figure 2). There were 2 complications in patients who underwent subsequent surgeries. An extension of a laminectomy into the thoracic zone resulted in a dural tear, and another patient who underwent a posterior cervical procedure after a previous lumbar decompression was left with persistent hand numbness postoperatively.

Four patients (16%) underwent a third procedure during the follow up period, all were patients with achondroplasia. The average time between the second and third procedures was 10.0 $\pm$ 6.6 months. Two patients underwent a revision, one underwent an extension of decompression with fusion, and one underwent a procedure at a non-contiguous anatomic zone. There were 2 complications, both in revision thoracolumbar decompressions—1 durotomy and 1 surgical site infection requiring irrigation and debridement.

Discussion

The findings in this study disputed our hypothesis. Specifically, contemporary, multidisciplinary, and coordinated perioperative care for this complex patient population has not led to a marked improvement in complication rates.[12,19] In our series, 48% of patients experienced a complication, a rate comparable to much older series.[4,9] Similarly, these patients were found to have a high rate of revision surgery, with 36% of the patients in our series requiring subsequent surgery over a five-year follow up period.

Prior studies show similar rates of complications as those seen in our series. In our population, dural tears were seen in 36% of cases, neurologic deficits in 12%, and infections in 8%. Similarly, in a series of 98 patients with achondroplasia undergoing laminectomy between 1970-2003, Ain et al reported a dural tear rate of 37%, a neurologic complication rate of 23%, and infection rate of 9%. This leads to the conclusion that certain complications, such as dural tears, may be secondary to intrinsic, non-modifiable risk factors. These include anatomic sequelae of the underlying dysplasia, such as severity or chronicity of congenital stenosis, increased lumbar lordosis, horizontal positioning of the sacrum, and increased thoracolumbar kyphosis, which may predispose the patient to complications.[20] High infection rates may also be attributable to patient-level factors, such as higher prevalence of increased visceral adiposity, especially considering that high infection rates have been reported after other musculoskeletal surgeries in this population.[10] Given that neurologic deficits may also rely more on other non-anatomical factors such as blood pressure management or anesthetic technique, it is possible that advances in these areas has helped to contribute to a lower incidence of neurologic deficit after spine surgery.[14]

One notable difference in our series is that we included patients with diastrophic dysplasia and spondyloepiphyseal dysplasia. While complication risk may vary across these different dysplasias, we are unable to draw any conclusions with regard to differences by underlying skeletal dysplasia diagnosis, due to our limited sample size. The majority of research on spondyloepiphyseal dysplasia has been focused on the treatment of C1-2 instability in children, with no case series reporting on results after thoracolumbar surgeries in adults.[2,21,22]

As with the surgical complication rate, the medical complication rate in our series is also comparable to that reported by prior studies. Both complications, a pulmonary embolus and a urinary tract infection, occurred in a single patient. While the low number of medical complications makes comparisons somewhat arbitrary, this figure is consistent with past literature. Vleggert and Eul, for example, reported 1 UTI out of 25 surgeries in achondroplasia,[4] and Ain et al reported a 3% rate of venous thromboembolic event (VTE).[9] Coordinated multidisciplinary initiatives have been shown to have a larger effect on medical complications compared to surgical complications.[19] However, given that medical complications were already low in this population, it is possible that the complication rate could not be lowered further, regardless the degree of intervention. Additionally, gathering a large sample size reflective of a significant reduction in complication rates is challenging, giving the rarity of skeletal dysplasias.

Table 2. Complication Rates in Series.

Complication	n	%
Durotomy	9	36%
Acute infection requiring irrigation and debridement	2	8%
Genitourinary	1	4%
Neurologic	3	12%
Thromboembolic	1	4%

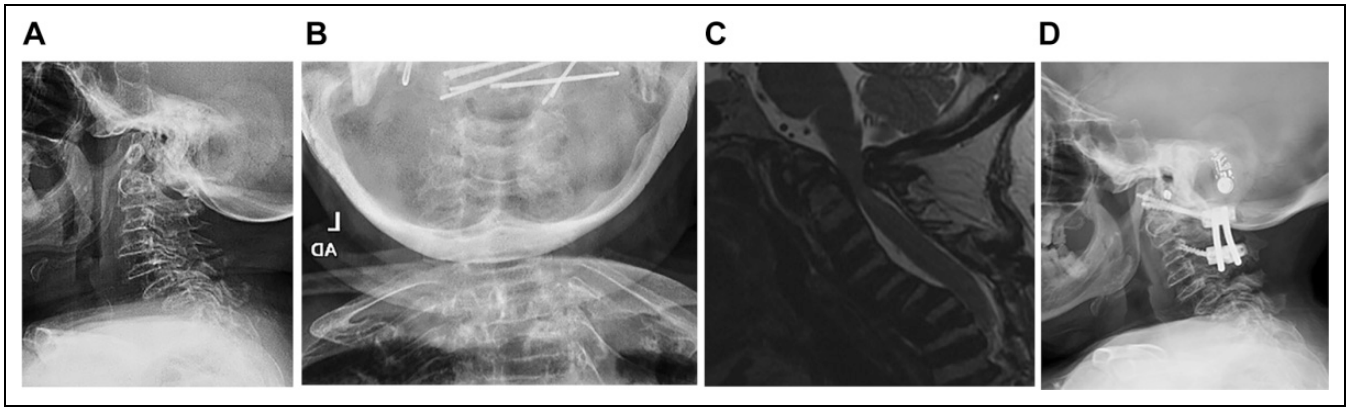

Figure 1. 64F with spondyloepiphyseal dysplasia with signs and symptoms consistent with cervical myelopathy. Imaging consistent with os odontoideum and atlantoaxial instability (Figure 1A-, 1B, 1C, flexion/extension views not shown). Patient underwent suboccipital and C1 decompression and C1-C3 posterior fusion. C2 was not instrumented due to a high riding vertebral artery on the left and a diminutive pedicle and pars on the right. In accordance with the preoperative plan, the patient was kept intubated postoperatively and extubated on POD1. A CT chest was performed on POD5 showing a large central pulmonary embolus within the right main pulmonary artery. An IVC filter was placed the same day, and the patient was started on therapeutic enoxaparin the next day. On POD8, the patient was started on antibiotics given dysuria and increased frequency in the setting of a positive urine culture. Two-year follow up shows no hardware displacement and a solid fusion (Figure 1D and E).

Table 3. Revision Procedures Included in Series.

Patient	Age at initial surgery	Skeletal Dysplasia	Initial Surgical Procedure	Indication for Second Surgery*	Second Surgery*	Revision/ Extension	Time between procedures (months)	Indication for Third Surgery	Third Surgery	Time between procedures (months)
1	60	Achondroplasia	PD L1-L5	Cervical myelopathy	ACDF C7-T1		1.0	Thoracic myelopathy	PDF T6-T10	1.0
4	64	Achondroplasia	PD L1-S1	Cervical myelopathy	ACDF C4-C7		7.3	Thoracic myelopathy	PDF T9-L1	9.7
7	15	Achondroplasia	PD T10-L3	Lumbar radiculopathy	Revision PD L2-3, PLD L3-5	Extension	6.7			
9	42	Achondroplasia	PD L1-S1	Cervical myeloradiculopathy	PDF C4-C7		6.4	Recurrent neurogenic claudication	Revision PD L3-L5	13.8
11	59	Diastrophic Dysplasia	PD T12-S1	Cauda equina syndrome	Revision PDF T7-L2	Revision/ Extension	0.4			
17	59	Achondroplasia	PD L2-S1, PF L3-L4	Lumbar radiculopathy	Revision PD L4-L5	Revision	16.3			
22	45	Achondroplasia	PD T11-S1	Bladder incontinence	PDF T10-L1, AF T12-L1	Extension	5.8			
23	13	Achondroplasia	PD L1-L3	Progressive deformity	Revision PDF T10-L4	Revision	10.2			
24	50	Achondroplasia	PD T11-S1	Lumbar radiculopathy	Revision PDF L1-L4	Revision	39.4	Pseudarthrosis, thoracic myelopathy	Revision PD T9-L2, revision/extension fusion T9-L4	15.8

*Excludes irrigation and debridements. *PD, posterior decompression; PDF; posterior decompression & fusion; PF, posterior fusion; ACDF, anterior cervical discectomy and fusion; ROH, removal of hardware.*

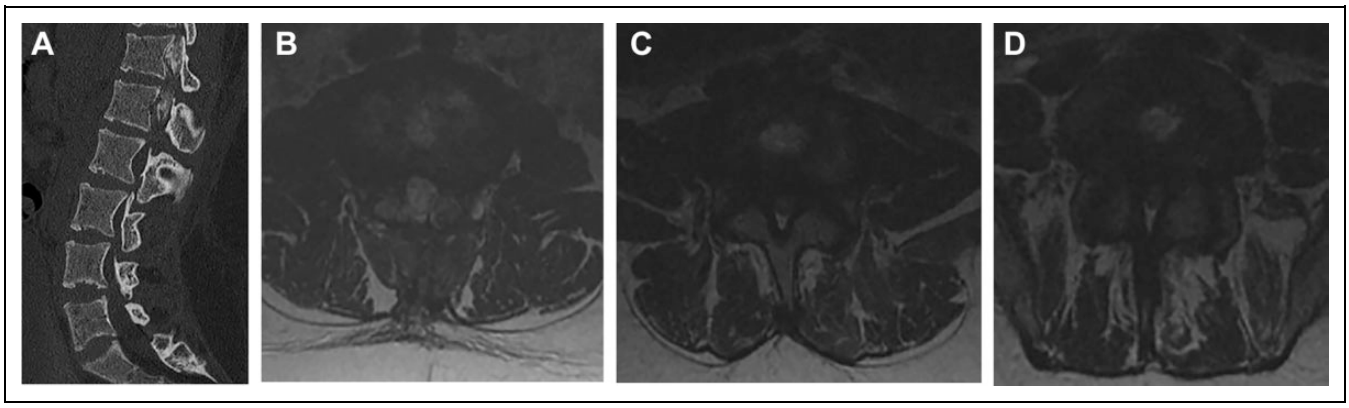

Figure 2. 18F with achondroplasia underwent bilateral tibial lengthening procedures with no immediate neurologic changes. However, the patient developed bilateral lower extremity weakness and buttocks numbness 3 days after the lengthening procedures. An urgent CT myelogram was performed, showing partial-complete dye blockage at T12-L2 levels (Figure 2A). The patient underwent an uncomplicated posterior decompression from T10-L3. Neurologically, she continued to improve postoperatively but plateaued with mild weakness in the bilateral lower extremities. An MRI was performed 4 months after the initial procedure showing continued moderate stenosis at L2-3 (Figure 2B) and severe stenosis at L3-4 (Figure 2C) and L4-5 (Figure 2D). The patient underwent revision decompression at L2-3 and decompression of L3-5, complicated by an unrepairable dural tear. One year after revision procedure, the patient exhibited improvements in distal right lower extremity strength only.

While medical complication rates are comparable to historical studies, this does not negate the importance of improving multidisciplinary perioperative care efforts. White et al. published a consensus of 19 to recommendations to mitigate known perioperative complications in this population. Formulated by a panel of 13 experienced specialists, including orthopaedic surgeons, anesthesiologists, geneticists, and others, these statements addressed airway management, positioning risks, and abnormalities in chest wall and body habitus.[14] Such initiatives may have affected other outcomes, including anesthetic complications, immediate post-operative care, patient satisfaction, discharge to home, follow up rates, etc; however, these outcomes that were not a focus of this study.

To our knowledge, this is the first series to specifically look at the frequency of subsequent spine surgery in an adult spinal dysplasia population. While few studies have specifically examined rates of subsequent surgery, in past case series, revision surgeries comprised between 25% to 29% of the initial patient sample.[4,9] In a mail survey of patients with achondroplasia, Carlisle et al found that 49% reported a subsequent spine surgery between 1 to 28 years after laminectomy.[8] Of note, this included surgeries performed at other centers, while our series only captured surgeries performed at the same institution, and thus likely underestimates the true rate of subsequent surgery. In our series, half of subsequent procedures required either a revision or extension of a previous decompression or fusion; all indications involved a return or worsening of neurologic symptoms. Due to the underlying pathology, many of these patients have disease across several levels and it can be difficult to judge which levels are contributing to the symptoms preoperatively. Additionally, aberrant anatomy (e.g. short pedicles) increases the risk for disease progression. Given the substantial rate of additional surgery for adjacent segment disease, particular care should be taken both preoperatively and intraoperatively to plan levels for a complete decompression and avoid unnecessary second surgeries.

Finally, the incidence of subsequent spine surgery at non-contiguous segments suggests that a thorough work-up of all spinal segments should be undertaken in the evaluation of skeletal dysplasia patients with neurologic complaints. For patients whose initial evaluation indicated the need for surgery at non-contiguous zones, multidisciplinary care programs can help facilitate the discussion of goals, preoperative medical testing, anesthetic requirements, and timing of procedures.[14]

There were several limitations of this analysis. First, utilizing only a single-institution series decreases heterogeneity of surgical technique and perioperative care, introduces selection bias and limits generalizability. Certain outcomes, such as the need for subsequent surgery, are more heavily influenced by this bias, as surgical indications are often subjective and dependent on joint surgeon-patient decision making. Selection bias was also introduced through incomplete charting of certain patients, given that 7 out of 39 patients from the initial cohort were excluded due to incomplete data. Second, we were unable to comment on differences in outcomes in patients with different types of skeletal dysplasia. Large cohorts of patients with rare diseases are often not available for study. This is especially true for adults, who typically lack access to coordinated care once they age out of pediatric hospitals. Comparing surgical outcomes between patients with different types of skeletal dysplasia will likely require a multi-institutional effort. In addition, our case series is somewhat heterogeneous, given the variety of procedures performed across different regions of the spine. Despite the variety of procedures performed, the indications and goals of surgery were largely similar; we believe makes the grouping of this cohort valid, especially given that historical complications have attributed to decompression rather than fusion.[9] Furthermore, we did not capture complications

or subsequent surgeries managed at outside institutions, which may lead to underestimations of our outcomes of interest. However, most patients who received surgery at our center receive all of their medical care through the multidisciplinary skeletal dysplasia center at our hospital, which provides a logistical incentive for patients to continue receiving all care at our institution. Finally, given that our perioperative medical care is administered through a specialized skeletal dysplasia clinic, the generalizability of certain findings may be limited, particularly with regard to rates of medical complications.

In conclusion, skeletal dysplasia patients undergoing spine surgery are at high risk for surgical complications, including dural tears or wound infections. Such complications are likely attributable to inherent patient factors, unaffected by contemporary advances in instrumentation or techniques. Surgeons must be keenly aware of the propensity for these complications both intraoperatively and when counseling patients. Furthermore, our series demonstrates the frequency of subsequent spine surgery in this population. Extensive preoperative planning, paying particular attention to the anatomic structures and levels responsible for the patient's symptoms, should help surgeons minimize the need for revision or extension surgery. Nevertheless, these patients may still be at higher risk for further stenosis and should be counseled according. Lastly, the evaluation of all spinal segments for potential or impending pathology is a must. Centralized, multidisciplinary care centers can coordinate the complex perioperative care process to mitigate risks for this unique patient population and optimize perioperative outcomes.

Declaration of Conflicting Interests

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IRB Exemption Statement

This study was approved for exemption of informed consent by the Institutional Review Board of the Hospital for Special Surgery as this study was an evaluation of patient records and documentation.

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