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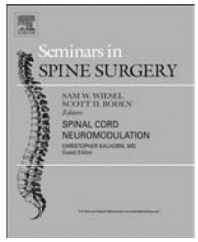

Revision spine deformity in pediatrics

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ABSTRACT

Pediatric spinal deformities requiring revision surgery are relatively uncommon to encounter and treat. These can, however, be challenging and require careful planning and execution. Functional impact of residual/ worsening spinal deformities should be carefully analyzed and the risks/ expected benefits should be discussed with patient/ families in detail.Spinal osteotomies may be a valuable tool and surgeons should be familiar/ comfortable with common osteotomy techniques for these cases.

> "Perfection is not attainable, but if we chase perfection we can catch excellence."
>
> -*Vince Lombardi*

1. Introduction

Revision surgeries in general are technically more demanding than the index procedures. They also have high risk of complications which can cause lower patient satisfaction. The number of deformity patients requiring revisions is expected to expand in coming years with global adoption of deformity correction procedures and increase in total number of primary deformity correction procedures. Advances in understanding of deformity biomechanics, surgical techniques and newer implants continue to improve our outcomes but it is still far from perfect.

Revision spine deformity surgery in children may be indicated for a variety of reasons including painful implant, neurological deficit, infection, implant failure, and a residual, new or worsening deformity.[1] Identification of factor leading to patient dissatisfaction or failure of index procedure can be challenging. Establishing relevance of radiological findings with respect to patient's symptoms can be even more challenging. Mere radiographic findings of residual / new deformity or pseudoarthrosis may not warrant surgery and may need to be evaluated in terms of functional impact.

1.1. *Incidence and risk factors*

The incidence of revision surgeries varies significantly in literature, depending on the instrumentation, follow-up period and underlying pathology & diagnosis, and has been reported to vary significantly from 0.9% to 75%.[2–10]

One of the largest follow up study for spinal deformity revisions was performed by Richards *et al.*[7] This study reviewed 1046 patients after adolescent idiopathic scoliosis (AIS) surgery and reported an overall revision rate of 13% (172 out of 1,046) over a period of 15 years. 77% of their patients had undergone posterior fusion, 22% anterior fusion and 1% had circumferential fusion. The most common reasons reported for revision were infections, symptomatic implant, and pseudoarthrosis. The posterior fusion cases had higher revision rates compared to anterior fusions (14.0% *vs.* 9.3%) possibly due to the use of single rod which has been shown to have higher failure rate.[7]

Uninstrumented spinal fusion and first generation spinal instrumentation techniques using Harrington rods were reported to have higher revision rates.[8,9,11] Dickson et al reported that 41% of patients who underwent Harrington

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instrumentation for scoliosis ended up having a broken rod in a minimum of 21-year follow-up.[8,11] Tamborino reported up to 37% of revision surgery after uninstrumented posterior spinal fusion and 16% with Harrington rod constructs.[9] Use of segmental spinal fixation has also decreased the rate of pseudoarthrosis between 0 and 4.1% in different studies.[11–13] Cook et al.[14] reported revision rate for different instrumentation system after AIS surgery. The overall rate of revision surgery after primary posterior instrumentation and fusion for idiopathic scoliosis was reported to be 19% in Cook et al. study on 182 patients.[14] Revision rate was reported to be 19% for Harrington system, 24% for C-D and 14 % for Isola instrumentation system at 9 years follow up.[14] Bago et al reported up to 25% of revision rate for Cotrel–Dubousset instrumentation in 10 year follow-up.[15] Vitale et al reported the rate of reoperation of 13% at 5 years follow up for a heterogeneous group of scoliosis patients.[16] In other studies 5.7% to 12.9% of patients required at least one revision procedure for infection, pseudoarthrosis or symptomatic implants in 2 year follow-up.[7,17]

The rate of revision surgery also varies with the type of deformity and underlying diagnosis. Lee et al reported 10-13% rate of revision surgery after regional instrumentation for Scheuermann's kyphosis.[18] Scoliosis surgery for diagnoses other than adolescent idiopathic scoliosis [19] and surgery on immature patients with open triradiate cartilage might have higher reoperation rate.[20] The immature patients with idiopathic scoliosis and open triradiate cartilages have greater risk of loss of correction after anterior instrumentation. Other causes include adding on levels proximally and distally leading to decompensation at the unfused lumbar curve.[20–23] Similarly, Gomez et al. also reported male gender and low preoperative Risser's score to be high risk factors for postoperative curve progression for all curve types.[24]

The rate of revision surgery has been reported to be 6.3% to 33% in congenital spinal deformities due to pseudoarthrosis and instrumentation complications.[25–28] This has been reported to be 53% after hemivertebrae resection in Bollini et al study, mostly for implant removal.[29] In case of neuromuscular scoliosis secondary to cerebral palsy and Marfan syndrome, 20% of the patient had to undergo revision surgery.[30,31] Thirteen percent of children with Duchenne muscular dystrophy required revision surgery in Alman et al. and Sengupta et al. studies.[32,33]

Patient with myelomeningocele also have higher risk for revision surgery to address curve progression (10%) or removal of instrumentation (up to 75%).[10,34]

Growth friendly instrumentation, surgery in metabolic and neuromuscular diseases as well as syndromic scoliosis has also been associated with high revision rate.[35–40]

It has been shown that bulkier instrumentation systems have higher revision rate compared to lower profile implants.[14] Kuklo et al. reviewed 1428 patients retrospectively after AIS surgery and concluded that antero-posterior as well as all pedicle screw constructs have lower revision rate compared to hooks and hybrid constructs.[41]

Miller et al concluded that patients with increased height or weight are at increased risk for curve progression in their analysis of 902 AIS fusion at 2 years follow up.[42] Lowest instrumented vertebrae in the thoracic region or high degree preoperative major curves are also noted to be high risk factors for coronal curve progression after surgery in this study.[42]

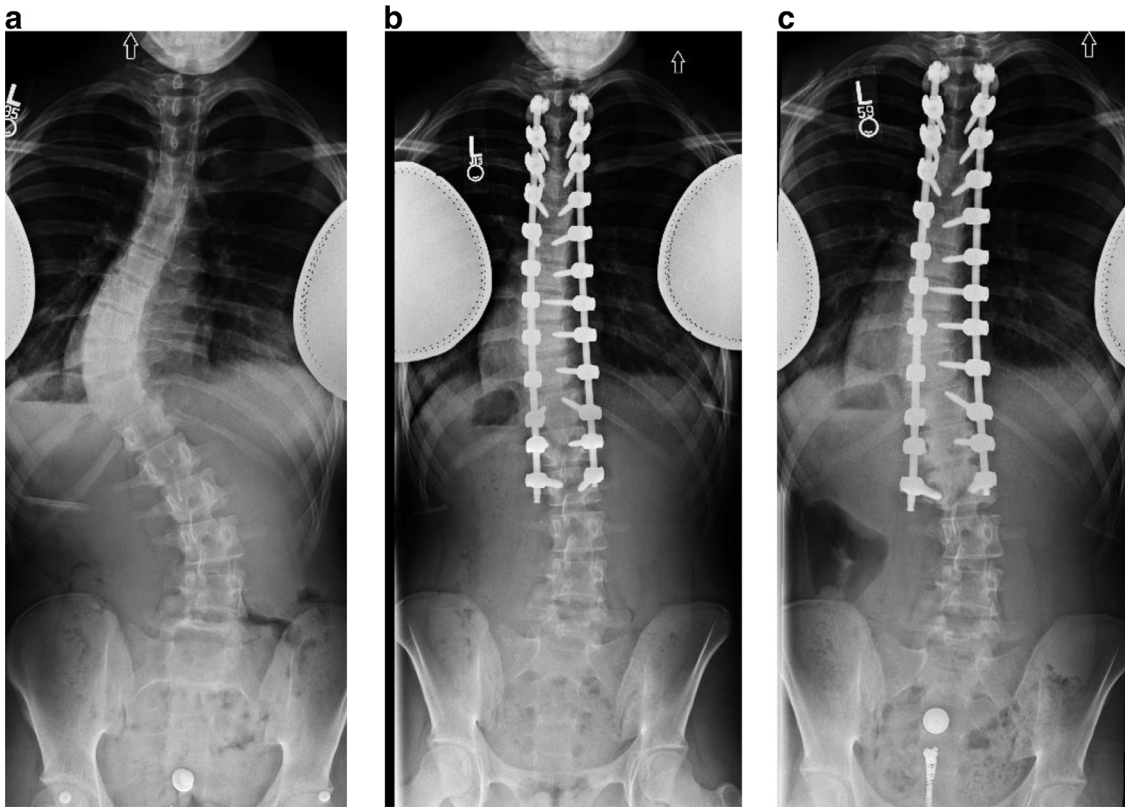

Fig. 1 – (a-c)- Asymptomatic distal level screw loosening with progression.

1.2. Clinical evaluation

Some of the common symptoms leading to revision surgeries include increased pain, deformity progression, neurological deficit and loss of function.[43] Pain is most common and might be due to acute (implant dislodgement) or gradual failure of the implant (Figure 1), pseudoarthrosis, or prominent instrumentation with irritation of the soft tissue, and also infection.[7,27,30,43,44]

Other symptoms leading to revision surgery include insufficient correction of the deformity, subsequent imbalance following the index surgery, additional levels of deformity in the coronal or sagittal plane around the operated levels (adding-on or junctional deformity) or late operative site pain.[11]

Detailed history of symptoms, thorough physical and neurological examination, activity/ functional assessment is paramount. The history should include the evolution of presenting symptoms, neurological symptoms, presence/ absence of any symptom-free period after index procedure and trauma/ sporting activities. Assessment of functional impact of the current symptoms is very important. The patient's desired activity level needs to be taken into account and appropriate counseling may be required to ensure relative overlap of surgeon and patient's expectations. Physical examination details should include prior incision site/ focal symptomatic site evaluation, neurological examination and overall Sagittal/ coronal plane balance. Careful evaluation of any leg length discrepancies, adjacent joints including hips and shoulders is required to rule out non-spinal pathologies.

1.3. Imaging

Plain Radiographs: Focal (cone down views) as well as full length spinal alignment images are recommended. Focal radiographs may help to identify any local/ regional fracture, instability or hardware failure. Full length imaging provides input for global alignment and is an essential tool for deformity correction planning. Stress imaging may also help to identify any evidence of instability. Traction, side bending or bolster images can be used to assess relative flexibility of the curves and can help to guide the surgical approach and osteotomies required.

CT/ CT Myelogram/ MRI: Three dimensional spinal imaging using CT scan is recommended to evaluate for prior instrumentation and fusion mass. Pedicle morphology and size can also be best assessed using CT scans. MRI is useful to evaluate the spinal canal and neural axis but its utility is relatively limited in cases with prior instrumentation. Alternatively, Myelograms may be added to CT imaging if spinal canal assessment is warranted in presence of prior hardware.

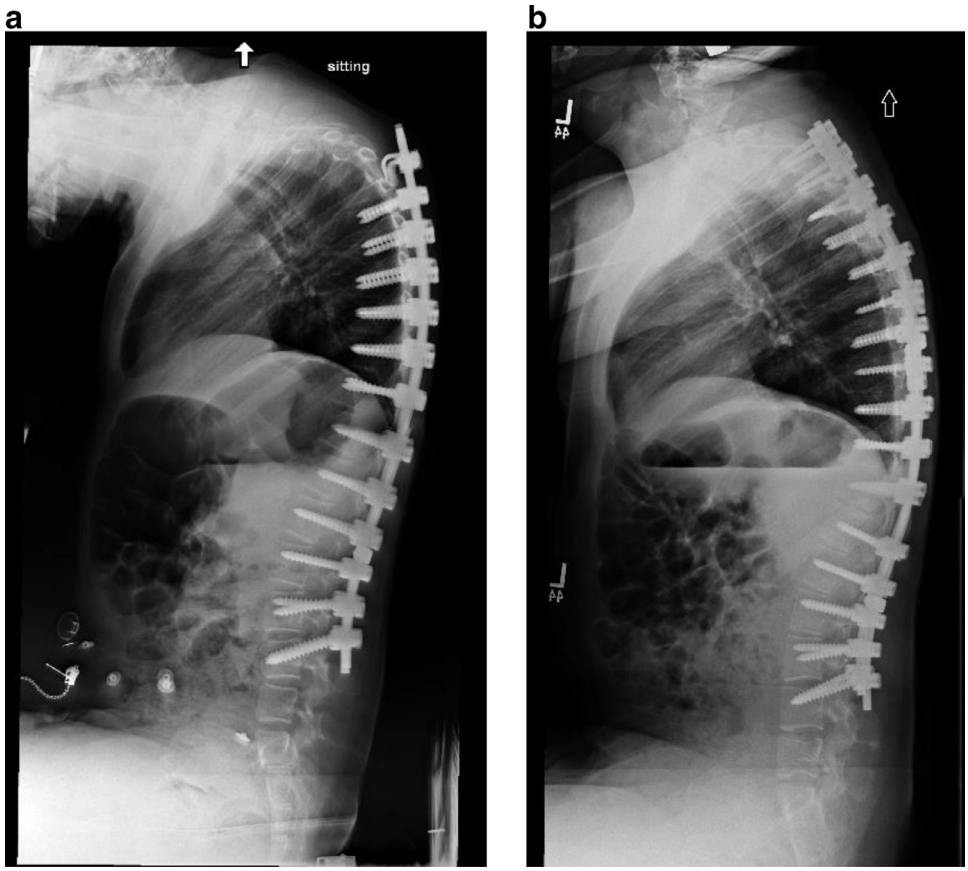

Fig. 2 – Proximal Junctional Kyphosis.
a- 8 years old boy with PJK (2 years after index surgery).
b- Revision spinal fusion for PJK.

Bone scan: Bone scans have limited utility for pediatric spinal deformities but may be considered for evaluation of infection, tumors and stress fractures.

2. Common diagnoses

2.1. *Proximal junctional kyphosis*

Proximal junctional kyphosis (PJK) is one of the most common recurrent deformity and is defined as- At least 10° of kyphosis between the lower end plate of the upper instrumented vertebrae and the upper endplate of the vertebrae 2 levels proximal to the upper instrumented vertebrae, and the Cobb angle must be at least 10° larger than the preoperative angulation at the segments.[45] (Figs. 2 & 3)

Following AIS surgery, the incidence of development of PJ K has been reported to be as high as 28%.[46] The proposed etiologies include: thoracoplasty, use of pedicle screws rather than hooks, obesity, failure to incorporate appropriate proximal vertebrae, lower Risser's grade, allograft use, and disruption of the ligamentum flavum at the proximal end of the construct.[46–48]

PJK can also be attributed to progression of the original deformity or degeneration of the segments proximal to the construct postoperatively. Spontaneous progression of the deformity above or below a healed spinal fusion might be due to improper selection of the upper and lower fusion and points.[43] This process can be accelerated by weak ligaments and muscles as well as suboptimal sagittal plane reconstruction at the proximal end of the construct. Junctional degeneration can be seen on x-ray by loss of disc height, increased angular deformity and degenerative disc disease at the levels proximal to the upper end of the construct.[43] PJK might be asymptomatic clinically without any effect on pain, patient's satisfaction and function score as well as self image. On the other hand, it can be painful, cosmetic concern or can cause imbalance/ feeling of falling forward in severe cases.[36]

For symptomatic cases of PJK, extension of the construct to 2-3 levels above the previous upper end of the construct with possible osteotomy, resection of the overgrown apical bone and anterior column reconstruction should be considered. A moderate correction of the kyphosis is the goal since the compensatory curve might be rigid and overcorrection might cause stress at proximal instrumentation with resultant screw pullout and development of recurrent kyphosis. The surgeon can lock the proximal screws to the rod and through the correction maneuver by cantilevering towards the fused segments.[19]

2.2. *Decompensation*

Scoliosis Research Society (SRS) describes balance as the static measurement of the alignment of the spine at any point in time while compensation is the active process of becoming balanced. Failure of achieve/ maintain balance of the body alignment is decompensation and may require surgical correction. Sagittal decompensation is defined as more than 5 cm of distance between the C7 plumb line and the posterior edge of the upper sacral endplate. Coronal decompensation is the progression of coronal deformity to the point that there is more than 2 cm of distance between C7 plumb line and the central sacral vertical line.[49] (Fig. 4)

Roberto et al. reported 28% of coronal curve progression (more than 10°) after spinal fusion for AIS. This was higher (54%) for patients with open triradiate cartilage, and selective thoracic fusion in Lenke 3 scoliosis.[50] It has been proven that sagittal balance is the most reliable predictor of clinical outcomes after deformity surgery and more than 4 cm of coronal imbalance is associated with higher pain and low functional score.[51]

More than 50% of coronal plane correction was associated with worsening sagittal balance at 2 year follow-up after AIS posterior fusion. Flat back is another complication of scoliosis surgery. If the thoracic spine is fused in hypokyphosis the lumbar spine will compensate with gradual hypo-lordosis to maintain the global sagittal balance. This can subsequently cause positive sagittal balance and increased pelvis tilt. Flat back deformity can cause pain, decreased function, and puts high stress on patient erector spina muscles.[45]

2.3. *Neurological deficit*

Global or focal neurologic deficits are rare in patients with spinal deformity correction. It has been reported to have an incidence of 0.35% in a National Inpatient Database (NIS) analysis of 35,600 pediatric deformity surgeries.[3] Similar analysis of large SRS database found an overall incidence of 1.0% (199 out of 19,360 patients). Neurological deficit complication varies from 0.8% (86 out of 11,227) in AIS to 2.0% (41 out of 2012) congenital cases in this SRS morbidity database analysis.[2] Progressive spinal deformity can happen as a result of myelopathic changes which is caused by the spinal cord being draped over the highly rotated and angulated curve.[43]

2.4. *Pseudoarthrosis*

Pseudarthrosis is the absence of bridging bone in between the fused segments. On radiographs a gap might be seen between the endplates of the vertebrae and the bone graft. Other signs include graft failure, radiolucency around the screws or implants, and finally implant failure.[49] Patients might have gradual onset of pain, progression of deformity, and failed instrumentation. There might be tenderness over the spine and painful range of motion in the area of original fusion.[43] Radiographs including flexion extension views, bone scan, and CT scan can be used for diagnosis.

2.5. *Crankshaft phenomenon*

Crankshaft phenomenon is more than 10° of coronal deformity progression with significant vertebral rotation. [49] This can happen in patients with several years of remaining growth who undergo posterior only fusion. Additional growth of the vertebral bodies causes overgrowth of the anterior column of the spine causing crankshaft deformity. [43]

2.6. *Definitive fusion after growth friendly system*

Failure of growth friendly systems to prevent progression or delayed worsening of deformity may warrant revision spinal

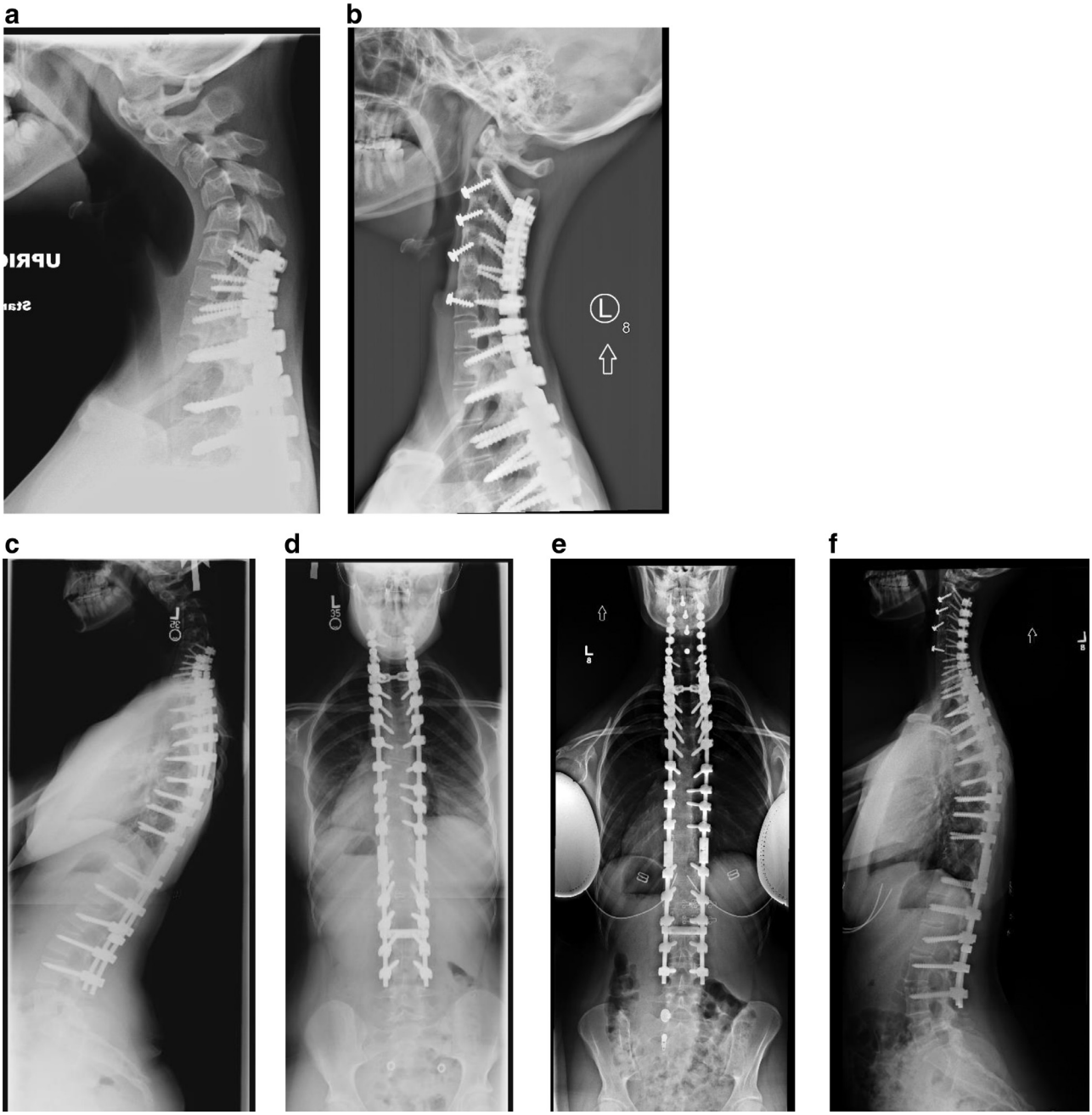

Fig. 3 – Proximal Junctional Kyphosis.

a & b- Cervical spine images showing PJK and post revision spinal fusion with extension of instrumentation
c & d- PJK as seen on full length spinal images
e & f- Post spinal instrumentation extension.

surgery. General principles of implant removal and spinal instrumentation apply in these cases. Staged procedures should be considered especially for very small patients. Poor anatomical landmarks, dysmorphic pedicles and extensive fusion may be encountered and CT imaging is helpful for planning. In case of spontaneous intervertebral fusion there might be a need for osteotomies. [19]

2.7. *Symptomatic implants*

Removal of existing hardware only, without reimplantation, may be indicated in case of prominent hardware. Complete or partial instrumentation removal may be an option for significantly symptomatic implants. Caution must be exercised to confirm the adequacy of fusion mass. Removal of the implant

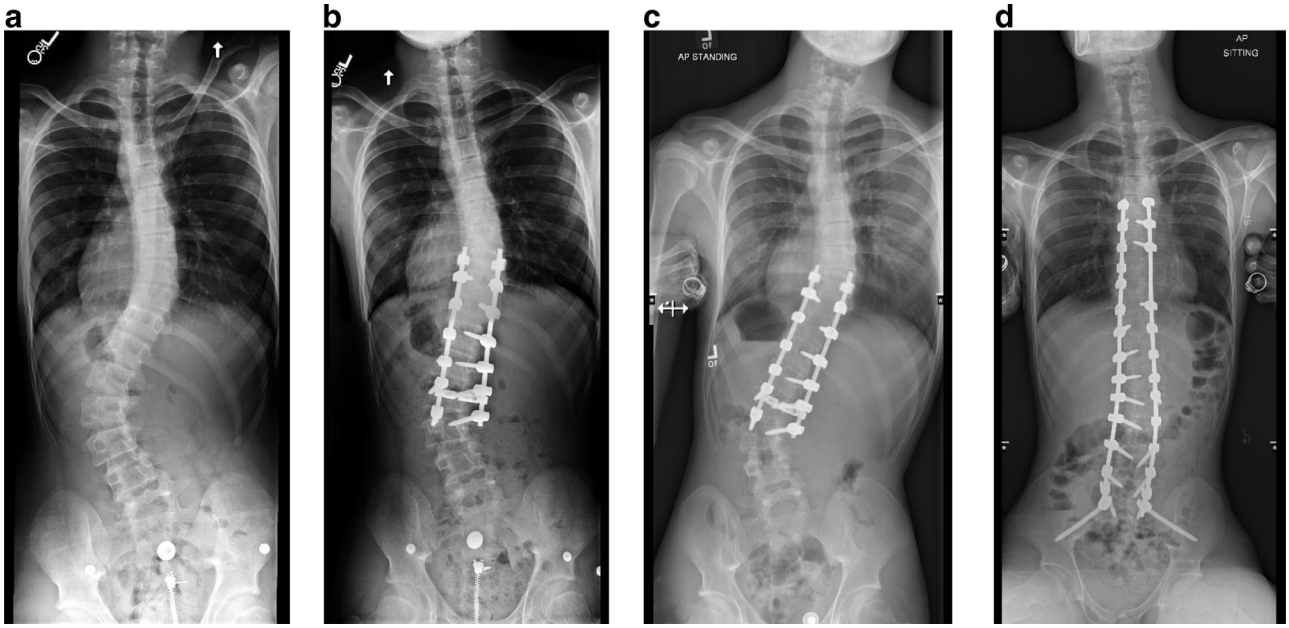

Fig. 4 – Decompensation with distal adding-on phenomenon.

a-Pre operative image
b-Post index spinal fusion
c-3 years after index spinal fusion with decompensation
d- Post revision spinal fusion for deformity correction.

or making defect in the previous fusion can be a stress riser and cause fracture in the fusion mass. [19] Other potential complication of implant removal is recurrence of deformity. Role of external brace wear after implant removal has not been substantiated. According to Cook et al. this problem is not seen often after removal of implants in immature patients since the late anterior column growth can buttress the spine against the post implants removal kyphosis.[14]

2.8. *Infection*

Early post-operative infection rate (within 90 days) is generally considered low (less than 1%) after spine fusion for idiopathic scoliosis, but has been reported to be as high as 9.7%.[1,12,15,52–54] Obesity with Body Mass Index (BMI) >95th percentile for age has been identified as high risk factor for infection in AIS patients.[54] Other reported high risk factors include the number of level fused, osteotomy levels and associated co-morbidities. The most common organisms were reported to be P. acnes, S. epidermidis, and Micrococcus varians in Richards and Emara study on 23 patients with deep wound infection after spinal fusion. They recommended irrigation and debridement and implant removal as a single stage surgery. There were no wound healing complications and no recurrence of infection.[55] The risk of infection is reported to be higher in neuromuscular scoliosis- 5.3-14%. The high risk factors for neuromuscular patients include incontinence, length of fusion, blood loss, hospital stay duration, obesity, malnutrition and pelvic fixation.[56]

If during the irrigation and debridement of a posterior spinal infection the tissues are viable, pink and clean, primary closure can be performed. However, if the tissues are not healthy or if the primary closure can't be performed due to excessive tension on the skin and soft tissue, the wound needs to be left open with the use of wound VAC. Vancomycin powder use in incision site after spinal surgery has been reported to reduce surgical site infection risk and should strongly be considered.[56–60]

In case of late infections (more than 3 months after surgery) treatment is controversial. Implant removal may be considered after surgical fusion confirmation. If implants are needed to be removed prior to fusion for resistant infection, reimplantation should be considered after infection resolution.[56] In the Hedequist et al.'s study on 26 cases they were not able to salvage any of the implants after a mean of 1.7 attempts. They removed the rods and debrided the supine and braced the patients however one fourth of the patient's needed re-instrumentation due to correction loss.[27]

Titanium is a more favorable implant in the presence of infection. In a study by Muschik et al. 7 out of 7 cases of infection were treated with single stage debridement and implant exchange with titanium instrumentation. They were able to eradicate the infection in all these 7 cases.[61] In another study mixing bone graft with gentamycin lowered there are infection rate from 15% to 4%.[62]

2.9. Surgical planning

Meticulous planning and careful attention to details is the key to the safe revision surgery. Discussing the case with a fellow surgeon especially when the case involves surgeon's own primary procedure is very helpful. Previous notes need to be reviewed which gives clues regarding the structural defects in the midline, blood loss, neural monitoring issues during the index procedure, medical problems during the surgery, and type of implant. In growing children the risk of growth cessation and resultant short trunk should be weighed again just the progression of the deformity. Preoperative lab work (erythrocyte sedimentation rate, C-reactive protein level, and peripheral blood cultures), combined gallium and bone scan, and aspiration of instrumentation can help identify infections.

Malnutrition is an important factor especially in very young and neuromuscular patients and may need to be treated prior to the procedure. Malnutrition can be worked up by measuring serum prealbumin level, blood transferrin level, and total lymphocyte count. Serum vitamin D, N-telopeptide, osteocalcin, and bone-specific alkaline phosphatase levels can be used to identify metabolic bone disease. If thoracotomy is in the plan, the patient may require a pediatric pulmonary consultation and pulmonary function test.[43] In unusual situations (congenital deformity) 3-D images or the physical model of the scoliosis can be helpful to guide the location of the osteotomies.

The surgeon should be aware of type of instrumentation used in the index procedure to be able to remove the implants if necessary and also plan for implant to implant connections. The extent of instrumentation is another major factor in planning. Anterior release can help reduce the need of major osteotomies and correction needed on posterior side. However, its utility is restricted because of relatively uncommon/ unfamiliar approach and risk of pulmonary complications.

Neuromonitoring can be extremely valuable tool to avoid devastating neurological complications and should be used wherever possible. Gavaret et al. reported a higher rate of neural monitoring signal change in revision cases as opposed to primary spinal fusions (19% in revision surgery vs. 11% in primary surgery) in 300 consecutive cases prospective study with no major post operative neurological deficit in either group.[63] Sagittal plane correction is another high risk factor for neuro monitoring change.[49]

Blood loss, as expected, is generally higher in revision surgeries.[62] A metaanalysis by Gill et al. showed that aminocaproic acid, tranexamic acid, and aprotinin decreased blood loss and the need for transfusions in spine surgery.[64] Use of aminocaproic acid, fresh-frozen plasma, cell-savers and other pharmacologic agents can also limit blood loss, which can sometimes determine the length of surgery the patient can tolerate.

2.10. Deformity correction tools

This section will cover the decision making and techniques of deformity correction especially some of the common osteotomies. Halo gravity traction can be very helpful in rigid deformities. It can be used pre-operatively or after anterior or posterior release (in between two stages for staged procedures).[65] In the second stage of surgery instrumentation and correction can be performed along with more aggressive osteotomies including pedicle subtraction osteotomy or vertebral column resection if necessary. For secondary curves after the index procedure the instrumentation need to cover the whole extent of the secondary curves.[1] Traction radiographs under general anesthesia have been described in the literature and found to be very helpful to assess the flexibility of the curved as opposed to supine side bending films.

Deformity correction using osteotomies is the most important armamentarium for revision cases and should be planned with absolute details. Important factors that can help to plan the osteotomies and deformity correction maneuvers include flexibility/ rigidity of the curve, overall alignment/ decompensation, focal vs. long sweeping deformities. For long sweeping and relative flexible curves multiple Smith-Peterson osteotomies might be the best strategy instead of PSO or VCR.[1,66,67] Focal, rigid and severe deformities might need more aggressive osteotomies including pedicle subtraction osteotomy (PSO) or vertebral column resection (VCR).[19,68]

2.11. Smith Peterson osteotomy

Posterior based osteotomies are valuable tools for deformity correction and can provide excellent correction if used at multiple levels in longer segment and flexible deformities. In this osteotomy the inferior facet, the spinous processes of the proximal vertebrae, supra and interspinous ligaments are removed. Subsequently ligamentum flavum is exposed and by using a finer rongeur a gentle midline bite can be taken from the ligamentum to expose the epidural fat. Kerrison rongeur can be used to remove the rest of the ligamentum flavum in oblique manner. This is important so to surgeon does not end up on the pedicle instead of the superior articular process of the distal vertebrae. Subsequently the superior articular process needs to be removed. Bleeding might be encountered during this stage since the resection is in the foramen. This can be controlled by the use of haemostatic agents or cottonoids.[19]

2.12. Pedicle subtraction osteotomy (PSO)

Pedicle subtraction osteotomy removes a wedge of bone from the anterior column of the spine. Subsequently the posterior portion can be hinged to close down to increase the lordosis. Pedicle subtraction osteotomy can be done in symmetric or asymmetric manner. The asymmetric PSO can be used to address the coronal imbalance. Pedicle subtraction osteotomy is used for rigid curves such as revision cases where the spine is fused.

2.13. Vertebral column resection (VCR)

Vertebral column resection includes complete vertebrae resection with the discs above and below. This osteotomy can be used for severe focal deformities to shorten and correct the rotational deformity.[19] A minimum of 3 levels, above and below the osteotomy level need to be included in instrumentation.[1] In the thoracic spine 4–6 cm of rib resection needs to be performed. In the lumbar spine, transverse processes need

to be removed. A posterolateral subperiosteal dissection can be done by Cobb elevators and bilateral retractors need to be placed on either side of the vertebrae to overlap in the front. A laminectomy will be performed at the level of the VCR. Proximal and distal levels are also recommended to be decompressed/ unroofed posteriorly to avoid any dural sac pinching at the time of deformity correction. There should be exposure from pedicle of the proximal vertebrae to the pedicle of the distal vertebrae. Nerve roots need to be protected during this procedure. Since the whole vertebrae is removed temporary rods spanning 2 vertebrae above and below need to be inserted on one side while the surgeon is working on the contralateral side. After identification of the pedicles, taps with increasing diameter will be introduced into the pedicle. The cancellous bone will be removed using curettes. The exiting nerve can be tied in the thoracic area however in the lumbar spine needs to be protected. After the removal of the cancellous bone the cortical shell can be removed using Leksell and Kerrison rongeurs.

The use of high-speed burr is imperative for sclerotic bone. In these situations large-bore burr (acorn tip) can help with quick bone removal. The resection needs to also be performed on the contralateral side. Posterior wall of the better break and be resected at the final stage of osteotomy using a down pushing curette since it protects the spinal cord during the removal of the vertebral body. The upper and lower end plates need to be prepared and dissected off cartilage. There is also a need for anterior column structural support (using bone or cage) in addition to posterior instrumentation when the osteotomy is done.[1,19]

In kyphoscoliotic deformities sagittal and coronal realignment can be safely done by the use of sequential bending in situ or multiple exchanging with better contoured rods.[1]

2.14. Summary

Revision surgeries for pediatric spinal deformities can be challenging. These are urgently indicated for very limited scenarios including major/ worsening neurological changes and very prominent implants. Majority of patients may continue to be safely observed to evaluate the impact of current symptoms/ diagnosis before undertaking major revision procedures. Revision surgeries tend to have unique requirements and need to be tailored to individual needs, even more than primary index procedures.

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