

Management of low back pain accompanying sagittal plane pathologies in children: Spondylolysis/spondylolisthesis and Scheuermann's disease

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Abstract

Background: Low back pain in childhood was underappreciated for a long time, but recent studies report higher prevalences, up to 70%. Two of the common causes are spondylolysis/spondylolisthesis and Scheuermann's disease. These disorders are relevant in a way they both cause significant back pain, and may disrupt the sagittal spinal balance.

Purpose: To present the current evidence on the diagnosis, natural history and treatment of these disorders with a special focus on sagittal spinal alignment.

Methods: This study is conducted as a literature review.

Results and Conclusions: Spondylolysis and low-grade spondylolisthesis have a benign course and are typically treated conservatively. When pars repair is indicated, pedicle screw-based techniques achieve more than 90% fusion with acceptable complication rates. High-grade spondylolisthesis, however, is frequently progressive. Surgical treatment involves fusion, which can be done in situ or after reduction. Reduction is useful for “unbalanced” patients to acquire sagittal spinopelvic balance, and it is important to distinguish these patients. Despite lowering the risk for pseudoarthrosis, reduction brings a risk for neurologic complications. With re-operation rates as high as 40%, these patients definitely require careful preoperative planning. Scheuermann's disease generally causes back pain in addition to cosmetic discomfort during adolescence. If the kyphosis is lower than 60°, symptoms typically resolve into adulthood with conservative measures only. However, it must be kept in mind that these patients may experience problems with physical performance and have a lower quality of life even when the problem seems to have “resolved”. Severe kyphosis and intractable back pain are the most frequently referred surgical indications, and surgery typically involves fusion. Proper utilization of osteotomies and proper selection of the upper and lower fusion levels are of utmost importance to prevent complications in these patients.

Keywords: Spondylolysis, spondylolisthesis, Scheuermann's disease, kyphosis, spinopelvic, sagittal balance, low back pain, adolescent

Introduction

Low back pain (LBP) is very common in the adult population and is pointed out as the leading cause of disability.¹ However, LBP in the pediatric population was believed to be rare, and, therefore, underappreciated for a long period. A 1989 study by Turner et al.² reported that less than 2% of pediatric orthopedic clinic admissions were related to LBP. Cross-sectional studies on larger populations revealed that this complaint is not so uncommon, reaching a life-time prevalence of more than 70%.³ Yearly incidence of pain episodes has been shown to increase during adolescence.⁴

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In younger children, constant LBP requiring medical attention is more likely to have an identifiable cause. However, older children mostly suffer from nonspecific LBP: more than 80% of adolescents with LBP were shown to have mechanical back pain.⁵ Underlying etiologies are various and may change significantly with the age of the patient.

This review focuses on two of the most common identifiable reasons of back pain that can also cause sagittal plane deformities in children and adolescents: spondylolysis/spondylolisthesis and Scheuermann's kyphosis.

Spondylolysis and spondylolisthesis

Epidemiology and natural history

Spondylolysis and isthmic spondylolisthesis in the pediatric population can be considered as different stages of the same pathologic process, which begins with a fracture in the pars interarticularis.⁶ High-intensity signal changes can be seen on magnetic-resonance imaging (MRI), and are generally considered and treated as precursor lesions.⁷ Natural history of spondylolysis, especially the initiating factors, is not very clear. It is believed to be a stress fracture seen in the pars interarticularis region as a result of repetitive microtrauma. Finite element models have shown that the pars region bears the highest amount of stress under loading, especially with extension and rotation of the spine.⁸ The lytic lesion does not occur in everyone under the same amount of stress, which implies the existence of certain predisposing factors, especially related to lumbopelvic anatomy. Upright position and ambulation seem to be strong contributors of this increased stress, considering that no nonambulatory or in utero or infantile cases have been reported for this condition. Genetic predisposition is hinted by family surveys, but more research is clearly needed.⁹

Spondylolysis in the pediatric population is most commonly seen at the L5 level (more than 80%), and L4 is second (5%–15%).¹⁰ Prevalence of this disorder may change significantly depending on age, ethnicity, gender, and level of activity. Studies reporting prevalence in the general pediatric population are relatively few and range between 3% and 7%.¹¹ Urrutia et al.¹² examined the computed tomography (CT) scans of 228 patients between ages 4 to 15 and reported a general prevalence of 3.5%. This number rises dramatically to 30% in symptomatic children with LBP as confirmed by MRI, making spondylolysis one of the most common reasons for LBP in this age group.¹³ Athletes have been shown to carry an increased risk for spondylolysis, and up to 50% of LBP in pediatric athletes have been attributed to this condition; but there is no consensus regarding which sport carries the highest risk.¹⁴

When the spondylolytic defect in the pars develops bilaterally, the vertebra may start to slip anteriorly,

hence called isthmic spondylolisthesis. According to the classification by Wiltse and Newman, there are five different types of spondylolisthesis: dysplastic, isthmic, degenerative, traumatic, and pathologic.¹⁵ Isthmic spondylolisthesis is the most commonly occurring type of spondylolisthesis in children and will be the focus of this review. The natural history and progression from spondylolysis to spondylolisthesis have been outlined by Fredrickson et al.¹⁶ in 1984, together with a 45-year follow-up of the same series by Beutler et al.¹⁷ In total, 68% of the first-grade students who were diagnosed with spondylolysis progressed on to spondylolisthesis. However, high-grade slip (>50% slip, grade III or IV according to Meyerding's classification)¹⁸ was not encountered in this series. More importantly, the authors observed that most of the slip progression happened during adolescence and slowed down in adulthood. These observations were confirmed by other studies.¹⁹

Clinical presentation and imaging

Many patients with spondylolysis and low-grade spondylolisthesis are asymptomatic and diagnosed incidentally. Patients usually present to the clinic with LBP that started insidiously. The pain may be central in the lumbar region or off to one side, and may radiate down to the buttocks and posterior thighs. True radicular pain and neurologic findings are very rare, and if present, the physician may look for a high grade (Meyerding's grade III or IV) or dysplastic spondylolisthesis. Clinical findings that are significantly related to early stage spondylolysis are identified as ≥ 4 weeks of LBP duration, interference with running, LBP starting with laterality and spinous process tenderness.²⁰ Every patient must be thoroughly inspected, and the gait must be observed. The most frequent inspection finding is lumbar hyperlordosis.²¹ Gait is usually normal in patients with spondylolysis and low-grade spondylolisthesis, but a shortened stride with flexion at the hips and knees may be observed, which might indicate a high-grade slip. Measurement of the popliteal angle can be used to confirm hamstring tightness²¹ in this scenario, and a complete lower extremity sensory and motor examination must be performed to look for possible nerve root irritations.

Several different sources of back pain in patients with spondylolysis and spondylolisthesis have been identified. The ligamentous soft tissue filling the pars defect was shown to have free nerve endings, and, therefore, can be stimulated to elicit pain.²² Due to local inflammatory factors, facet joints and radicular irritation can also become significant sources of pain. When high-grade slippage ensues, increasing instability is believed to become the main generator of pain.

Direct radiography is the most useful initial imaging modality in the evaluation of a child with LBP. Standing anterior–posterior (AP) and lateral radiographs are taken, and when a pars defect is suspected, supine oblique views

are considered to have better diagnostic value. Beck et al.²³ questioned the value of four-view radiographic series and found that standard two-view series had a sensitivity of 0.59 and a specificity of 0.96, and additional oblique views did not show significant benefit.

If a diagnosis other than nonspecific LBP is suspected, advanced imaging modalities are used. In addition to CT and MRI, single-photon emission computed tomography (SPECT) and bone scans can also be used. The Scoliosis Research Society (SRS) Evidence-Based Medicine Committee reviewed the current evidence in the literature regarding the use of these advanced imaging techniques when investigating spondylolysis.²⁴ CT is considered to have higher sensitivity and lower false-negative rates when compared with MRI, and, therefore, considered as the gold-standard diagnostic modality for this disorder.²⁴ It is also useful for following bony union; however, follow-up with CT scans causes an excessive amount of cumulative radiation and is not advised. MRI has the advantages of detecting early stress reactions in the pars which may be missed by CT and avoiding radiation exposure completely, and it is being used more and more frequently as the advanced imaging modality of choice, with up to 92% sensitivity in detecting pars lesions.^{21,25}

SPECT is more sensitive than bone scans and plain radiography. When compared with CT, it has the advantage of identifying stress reactions of the pars interarticularis without overt lysis and can help distinguish acute lysis from chronic nonunion.²⁶ Planar bone scans are rarely used nowadays because of the high false-negative and false-positive rates reported.²⁴ However, it is more sensitive than radiography and can distinguish between acute and chronic lesions, so it may still have a limited role in the clinical setting.²¹

There are several radiographic measurements used for the classification and assessment of spondylolisthesis. Meyerding's classification is the most widely used and measures the degree of slippage on standing neutral lateral radiographs.¹⁸ The slip is divided into the following categories: grade I: 0%–25%; grade II: 25%–50%; grade III: 50%–75%; grade IV: 75%–100%; and grade V: >100%, also called spondyloptosis. As modified by Dubousset, the slip angle (or lumbosacral angle) is measured as the angle between the line drawn along the posterior border of S1 and the line drawn along the superior endplate of L5.²⁷ The spinal deformity study group (SDSG) also devised a classification system for spondylolisthesis which takes into account slip grade, sacropelvic parameters, and global balance. This classification system is hard to utilize in the clinical setting and has low interobserver agreement due to its complexity.²⁸

Sagittal spinal balance and spinopelvic parameters are also important factors in both understanding the pathophysiology and evaluation of spondylolysis and spondylolisthesis. Hanson et al.²⁹ examined the relationship

between pelvic incidence (PI) and spondylolisthesis, and found that mean PI of the group with a slip was significantly higher than the controls. PI was also found to be positively correlated with slip grade. On a finite element model, increased stresses at the L5-S1 junction were observed with higher PI values.³⁰ Despite the strong relation between spondylolisthesis and PI, its predictive value on the progression of the slip is questionable and was not confirmed.³¹

Vialle et al.³² studied the effects of spondylolisthesis on global sagittal balance of the spine by comparing 244 patients with L5-S1 isthmic spondylolisthesis to 300 healthy controls. They concluded that despite an increased PI and a disturbed lumbosacral anatomy, global sagittal alignment is relatively well preserved by compensatory movements such as pelvic retroversion, increased lumbar lordosis (LL), and decreased thoracic kyphosis (TK). In contrary, Hresko et al.³³ identified two different subgroups among patients with spondylolisthesis: the “balanced” group had similar pelvic tilt (PT) and sacral slope (SS) measurements when compared with controls despite an increased PI, and the “unbalanced” group had a significantly retroverted pelvis with a high PT and low SS. By further studies, this “unbalanced” group was shown to be prone to positive sagittal imbalance.³⁴ These imbalanced patients constitute type 6 of the SDSG classification.²⁸ Identifying these patients may be important in deciding whether or not to reduce the deformity when surgery is planned.

Treatment options

Most of the patients with spondylolysis/spondylolisthesis are successfully treated by nonoperative methods. Surgical treatment is reserved for patients with high-grade spondylolisthesis or when nonoperative treatment modalities fail to achieve symptom resolution.

Nonoperative treatment may include activity restriction, rest, nonsteroidal anti-inflammatory medications, physical therapy, and bracing. Restriction of the offending activity is the mainstay of treatment, but the duration varies between 6 weeks and 6 months. Physical therapy may or may not be included in the treatment, specifically core strengthening, flexion-based lumbar exercises, and hamstring stretching.³⁵ Facet joint and epidural injections are also viable options to control refractory symptoms.³⁶

Although not supported by strong evidence, bracing is frequently used by physicians: 60% of the surgeons preferred to treat spondylolysis in adolescents with a brace in the sports medicine society.³⁷ Rigid thoracolumbar or lumbosacral orthoses have been reported extensively in the literature, but a recent study by Virkki et al.³⁸ on children with acute spondylolysis revealed that elastic lumbar support was not inferior to a rigid thoracolumbar orthosis. SRS-24 scores and bony union rates were similar

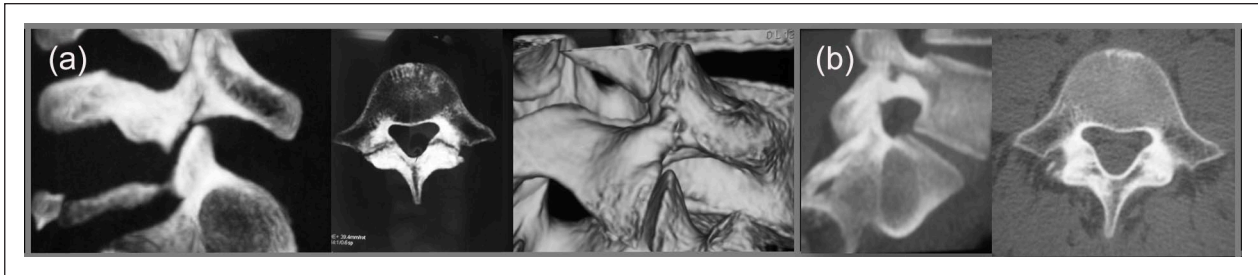


Figure 1. (a) L5 spondylolysis diagnosed by CT in a 12-year-old male patient. (b) After 6 months of conservative treatment with activity restriction, nonsteroidal anti-inflammatory medications and a soft brace, bony union, and total symptom resolution were achieved.

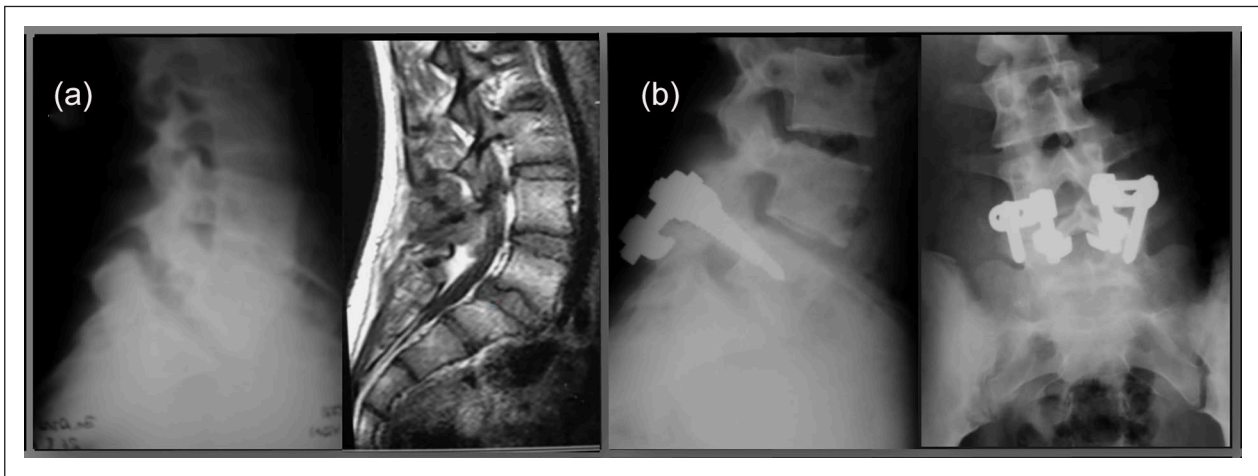


Figure 2. (a) L5 spondylolysis diagnosed in a 14-year-old female national team volleyball player. (b) Surgical treatment was indicated after 6 months of conservative treatment failed. Pedicle screw-based pars repair was performed with success, and the patient continued national team level sports activities afterward.

(69% for rigid bracing and 60% for elastic support, $p=0.785$). There is no strong evidence in the literature to prove that a brace can effectively restrict vertebral motion or unload the spine, but mostly it functions indirectly by limiting the excessive physical activities of the patient.

Nonoperative management of spondylolysis in children has a high success rate for symptomatic resolution (Figure 1). Young athletes carry a higher risk for spondylolysis, so early detection and treatment initiation is of utmost importance. In a recent study by Choi et al.,³⁹ 201 adolescent athletes with spondylolysis were treated conservatively (activity restriction, thoracolumbosacral orthosis, and bone stimulators) and 98% of the patients successfully returned to sports or similar preinjury activity levels. Eighteen percentage of them required facet or epidural injections before full recovery, and only one patient required surgery. Patients were followed up with CT scans and it must be noted that only 49.8% achieved bony union, which clearly demonstrates that bony union is not necessary for a favorable outcome. When treatment begins earlier in the disease process, bony union may be more likely. Sakai et al.⁴⁰ reported that 100% and 93.8%

of the lytic lesions healed when treatment was started in the very early and early stages, respectively.

Surgical treatment of spondylolysis is indicated when a nonoperative trial of 6 months fails to achieve symptomatic relief. Surgical treatment aims to repair the lytic pars region, and most frequently reported direct pars repair techniques include fixation with pedicle hook and screws, segmental wires (Scott method), lag screws through the fracture (Buck method), hook screws (Morscher method),⁴¹ and pedicle screw rod constructs (Figure 2). Mohammed et al.⁴² compared different repair techniques with a meta-analysis, and the reported pooled fusion rates were 83.5%, 81.6%, 77.7%, and 90.2% for Buck, Scott, Morscher, and pedicle screw-based repairs, respectively. In addition to the highest fusion rates, pedicle screw-based techniques also had a complication rate of 12.8%, which was significantly lower than the other groups. Low-grade spondylolisthesis can also be treated by similar methods. Direct repair techniques are frequently used to preserve motion segments, but it must be kept in mind that fusion of the affected segments is also a viable option with good results (Figure 3). Schlenzka et al.⁴³ compared the long-term results of segmental fusion

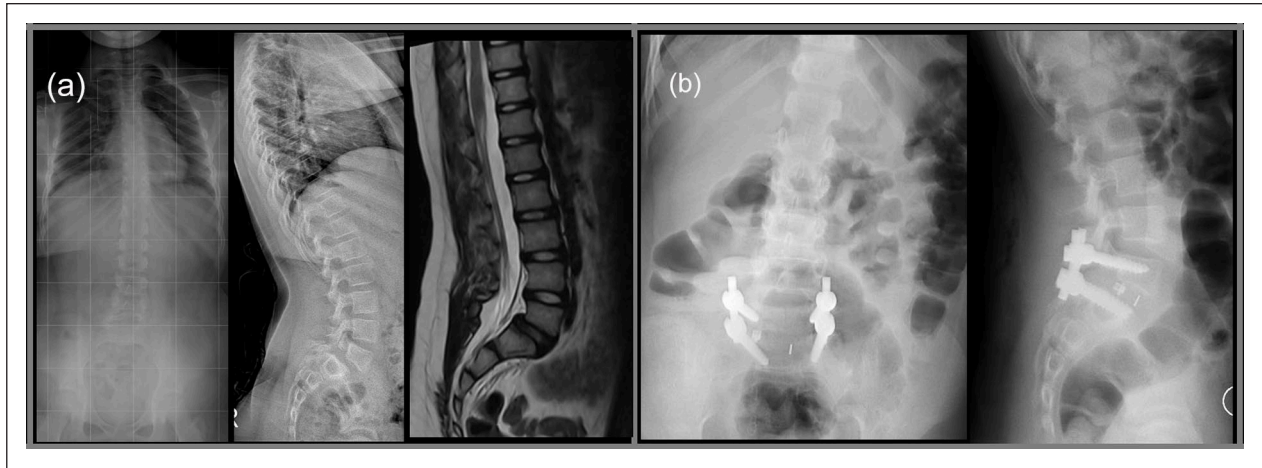


Figure 3. (a) Grade 2 L5-S1 spondylolisthesis in an 11-year-old female patient. (b) Circumferential fusion was performed including a transforaminal lumbar interbody fusion (TLIF), and total symptomatic resolution was achieved.

and Scott repair on patients with spondylolysis or low-grade spondylolisthesis. They reported that the fusion group had better Oswestry disability index (ODI) and SRS questionnaire scores. In addition, repair did not protect the olisthetic disk from degeneration.

High-grade spondylolisthesis is more frequently treated by operative methods, but conservative treatment can be initially offered to the patient if no neurologic deficits or cauda equina symptoms are present. In the case of a high-grade slip, Dubousset's lumbosacral angle $<90^\circ$ and sacral doming are other relative indications for surgical treatment. The debate in the literature focuses on the possible advantages and indications for reduction before fusion, and the type of fusion. In a very recent meta-analysis by Koucheiki et al.,⁴⁴ results of reduction and fusion were compared with in situ fusion. Patients undergoing reduction and fusion had a lower risk for pseudoarthrosis (risk ratio=0.51; 95% confidence interval=[0.26, 0.99]). Reduction was pointed out to have higher neurologic complications by many studies, and this meta-analysis noted that 5.5% of patients who underwent reduction had permanent neurologic complications compared with none in the in situ group. However, the difference between neurologic complications was not statistically significant. To reduce the risk for neurologic complications, osteotomies like the sacral dome resection have been suggested.⁴⁵ This technique shortens the lumbosacral region and prevents overlengthening of the nerve roots (Figure 4). Re-operation rates may also be disturbingly high after surgical treatment for high-grade spondylolisthesis: Nielsen et al.⁴⁶ reported on 50 patients that after a mean 2.1 years after index surgery, 40% required re-operations. It must be noted that partial reduction was performed in most of the cohort, as can be seen by the mean preoperative slip percentage of 61% dropping to 30% postoperatively. Implant failure was the most frequent re-operation indication, followed by persistent radiculopathy, infection, and back pain. Attention

must be paid to the SDSG classification and the difference between balance-unbalanced cases to assess the need for a reduction.^{28,33} Restoring the spinopelvic balance is gaining more and more emphasis every day, and the surgeon must find the correct method by weighing risks against re-alignment requirements for each patient.

Circumferential fusion has been compared with anterior or posterior-only fusion strategies for high-grade spondylolisthesis. Molinari et al.⁴⁷ reported 39% nonunion rate for posterior-only fusion when compared with none in the circumferential fusion. Circumferential fusion also achieved the best clinical outcomes after a mean follow-up of 17.3 years, when compared with posterolateral and anterior fusion.⁴⁸ Nielsen et al.⁴⁶ reported that the risk for re-operation did not change when anterior interbody fusion was added to a posterior fusion. The evidence in the literature seems to slightly favor circumferential fusion, but surgeon preference—experience is perhaps still the most important factor.

Summary—spondylolysis/spondylolisthesis

Spondylolysis and spondylolisthesis are common causes of back pain among children. As with any stress fracture, this pathological process runs an indolent course, and higher clinical suspicion is necessary for timely diagnosis. Advanced imaging modalities, especially MRI, are being used with increased frequency and allow for diagnosis as early as the prelytic phase.

The natural course of spondylolysis and even low-grade spondylolisthesis typically involves symptom resolution with conservative measures, but if surgery is warranted, pedicle screw-based techniques give the best results as indicated by current literature. High-grade slipage, however, requires a much different approach and a low threshold for surgical interventions. Understanding the differences between balanced and unbalanced

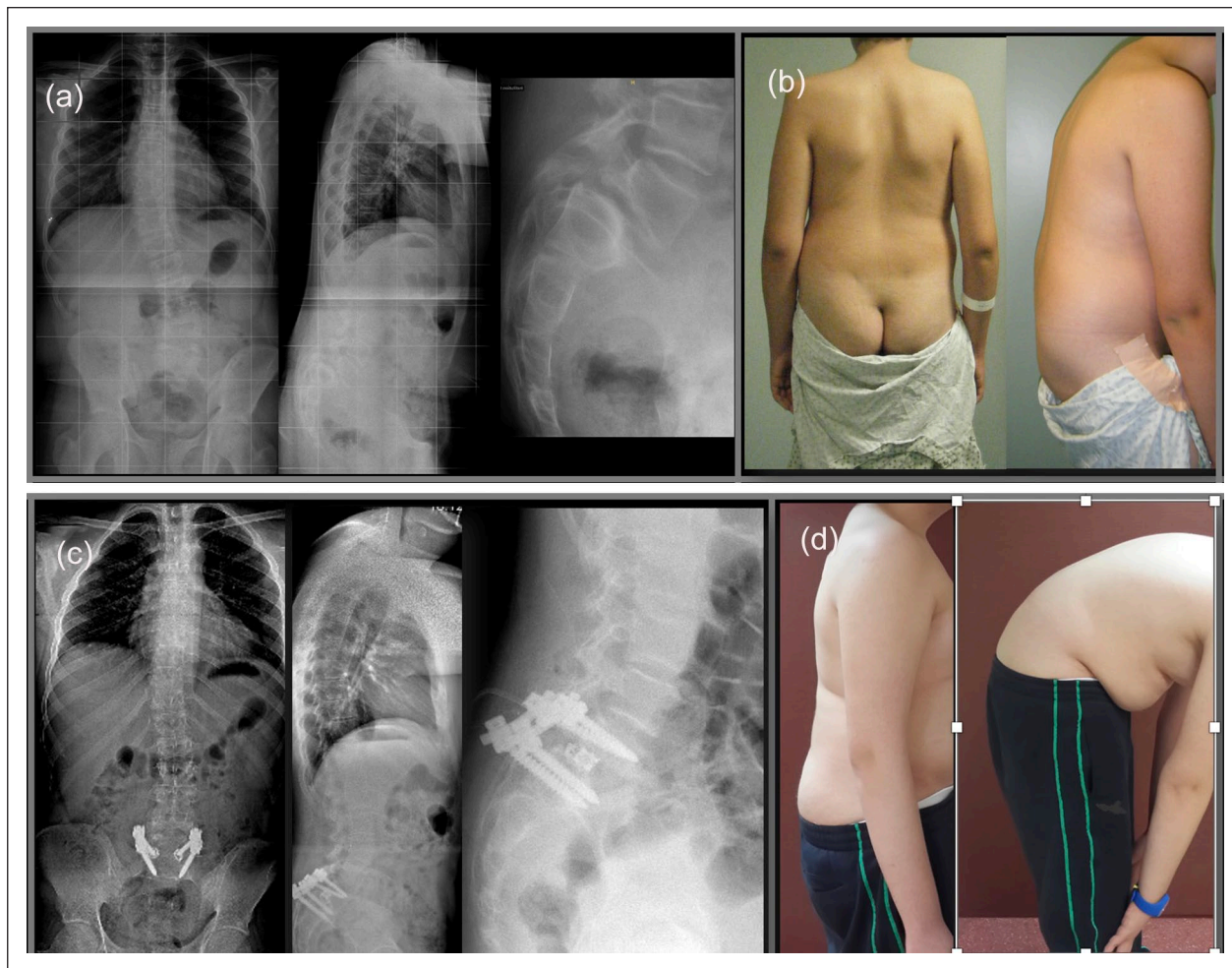


Figure 4. Radiographs (a) and clinical photographs (b) of a 16-year-old female patient with high-grade spondylolisthesis. Postoperative radiographs (c) and clinical photographs (d) have been shown, and the patient has been treated with a sacral dome resection followed by reduction and circumferential fusion including TLIF.

spondylolisthesis is the key to assess the need for reduction, and, therefore, restoring proper sagittal alignment.

Scheuermann's disease

Overview

Scheuermann's disease (SD) was first described by Scheuermann in 1921 as a painful, fixed, dorsal juvenile kyphosis of the spine characterized by defects in the vertebral endplate and vertebral wedging.⁴⁹ The overall prevalence of (SD) is 2.8% (ranging from 0.4% to 8%) and the male-to-female ratio is close to 2:1.⁵⁰ Diagnosis is usually suggested in clinical examination and further confirmed by radiographs. Widely accepted radiographic criteria of three adjacent wedged vertebrae angled by at least 5° were proposed by Sorensen;⁵¹ however, some authors suggest that one wedged vertebra is sufficient to conclude a diagnosis of SD if associated with irregular vertebral endplates.

Schmorl's nodes are herniations of intervertebral disk nuclear material through the adjacent endplates, and are frequently seen in SD. They were first described by Schmorl in 1927.⁵² The pathogenesis of these nodes is still unknown. They have also been found in up to 76% of the general population. They were found with a predominance in males, more frequently in the lower than in the upper vertebral end plate. They were also more common and more severe in the thoracolumbar than in the lower lumbar region.⁵³

There are two types of SD: type I "Classic" SD is the most common type, affecting the area between the seventh and ninth thoracic vertebrae (Figure 5), and type II or "atypical, lumbar" SD, which was first described by Blumenthal et al.,⁵⁴ is less common, affecting the lower thoracic spine (also known as the thoracolumbar spine) and sometimes the lumbar spine (Figure 6). The pronounced area, or apex, of the curve is between the 10th and 12th thoracic vertebrae.

The true etiology of SD remains unknown. It is thought to be caused by a genetic predisposition combined with mechanical stress on the vertebral endplates.⁵⁵ Osteonecrosis of the ring apophysis, loss of disk height, and vertebral morphology by the herniated disk materials and local vertebral osteoporosis have all been proposed by different researchers, but were not supported by the

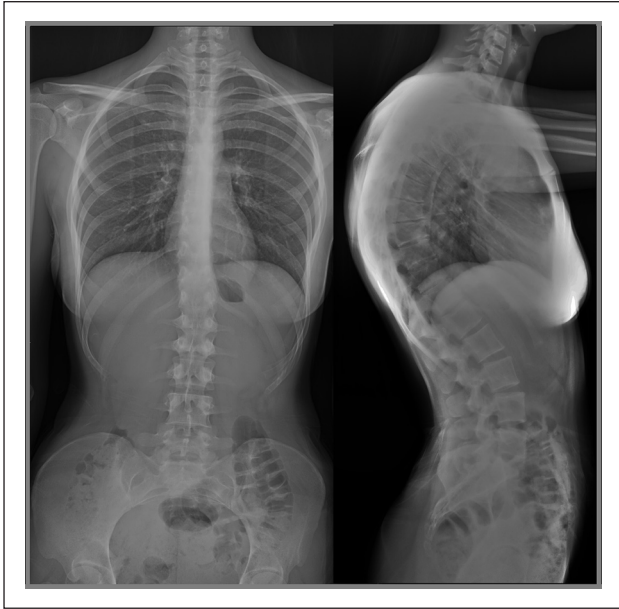


Figure 5. Radiographs of a 19-year-old female patient with type I SD. The thoracic kyphosis measures 78°, with an apex at T8.

following studies.⁵⁶ On a series of patients with type II SD, Greene et al.⁵⁷ suggested that excessive loading of the immature spine particularly in a preflexed posture may be responsible for the localized vertebral changes and subsequent back pain, thus pointing out to a more mechanical/traumatic basis for this subgroup of patients with lumbar/thoracolumbar disease. As a consequence, this disorder is commonly found in teenagers who are involved in strenuous activities such as weight lifting, rowing, and gymnastics. The vertebral abnormalities described were not found to be progressive.⁵⁷

Unfortunately, diagnosis of SD is often delayed, as it is misattributed to “poor posture.”

Posture and spinal alignment in SD

Researchers found patients with SD to be taller and heavier than healthy controls.⁵⁸ It has been proposed that the increased weight and height of these patients may be a result of other disturbances (i.e. hormonal), which may also play a crucial role in the pathogenesis of SD.⁵⁹ SD is a condition that has the potential to disrupt the spinal alignment in the sagittal plane. Thoracic hyperkyphosis, and the increased compensatory lordosis in the cervical and lumbar regions are not the only sagittal plane changes that are seen in this disorder. Spinopelvic relations have been analyzed in detail by Jiang et al.⁶⁰ Their findings revealed a significantly lower PI in young patients with thoracic and thoracolumbar SD compared with age-matched normal controls.⁶⁰ The authors indicate that patients with different

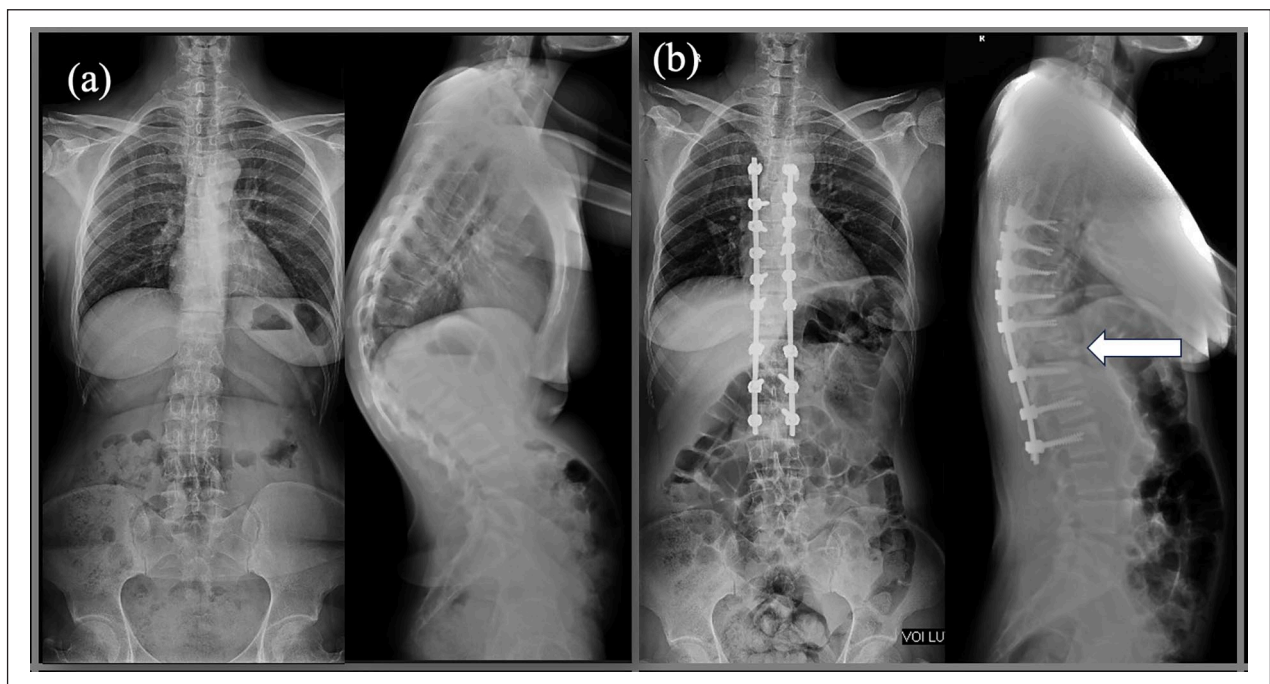


Figure 6. (a) Radiographs of an adult patient with neglected type II SD. The thoracic kyphosis measures 77°, with an apex at T11. Preoperative bolster radiographs to assess curve flexibility showed significant rigidity, and a pedicle subtraction osteotomy was performed (arrow) at the apex (T11) to correct the deformity (b).

curve patterns (thoracic and thoracolumbar disease) could have distinct compensatory mechanisms contributing to keep the sagittal balance.

Mostly overlooked, cervical spine may also be affected in patients with SD. Disease type (as either thoracic or thoracolumbar) affects the magnitude of cervical pathologies that may be expected.⁶¹ C0-2 lordosis was shown to be similar between the two disease types, but C2-7 lordosis was significantly greater in patients with thoracic disease, which indicates that compensation happens mostly at the subaxial region. T1 slope was also greater in patients with thoracic disease. Cranial radiological parameters (including C0 angle, C0-C2 angle, and cranial offset) and cervical sagittal vertical axis (cSVA) were comparable between the groups, and as indicated by this, position of the head center of gravity did not depend on type of the disorder, but more affected by the global postural compensatory mechanisms.⁶¹

Natural history

The natural history of SD tends to be benign. In patients with a kyphosis smaller than 60°, good clinical outcomes can be expected.⁶² In mild cases, the deformity may resolve spontaneously but in severe cases, treatment is usually necessary. Murray et al.⁶² reported on the natural history of this disorder on 67 patients who had a mean kyphosis of 71°, after an average follow-up of 32 years. They reported that the patients with SD had more intense back pain, jobs that tended to have lower requirements for activity, less range of motion of extension of the trunk and less-strong extension of the trunk, and different localization of the pain. However, there was no significant difference between patients and control subjects in terms of education, days absent from work due to LBP, presence of numbness in lower extremities, use of medication for back pain, or level of recreational activities. Both the patients and control subjects demonstrated the same levels of education as far as physical appearance is concerned. Patients with less than 100° of kyphosis had normal average lung functions. Those with greater than 100° of kyphosis with an apex between first to eighth thoracic segments had restrictive lung disease. In the study by Ristolainen et al.,⁶³ patients with SD more commonly reported back pain compared with healthy controls. The authors also found reduced quality of life when compared with healthy subjects. Although the difference was statistically significant, it was not immediately apparent in the patients' clinical symptoms. Patients with SD have reported lower self-esteem and poorer self-reported health, in addition to difficulties in performing activities of daily life, such as walking upstairs or carrying a load. As the authors suggested, these can be due to the effects of other problems like heart disease. In the described group, TK did not exceed 58°, but the patients still reported significant back pain. In addition to the changes related to SD in the thoracolumbar spine,

patients displayed secondary degenerative changes in the lumbar spine, which may be due to the compensatory increased LL.⁵⁸ In another study by Ristolainen et al.,⁶³ the rate of radiographic deformity progression, its mechanism, and the associations between kyphotic progression and clinical outcome in 46-year follow-up have been reported. They concluded that the kyphosis progressed more than 5° in two-thirds of the patients during a mean follow-up period of 46 years. Wedging of the affected vertebrae increased significantly over time but no correlation existed between the extent of kyphosis progression and function at final follow-up.⁶³ Garrido et al.⁶⁴ reported progression of kyphosis in untreated patients, from a mean 66° at skeletal maturity to 78° after a follow-up of 27 years. Long-term progression of untreated SD was calculated as 0.45°/year.⁶⁴ Neurologic complications have also been reported in a small number of patients, but as expected, they are only seen in severe kyphosis.⁶⁴

Characteristics of pain in SD

SD is a spinal deformity that is known to cause pain, especially in the lower back region. Many adolescents initially remain asymptomatic and may first present with poor posture and kyphotic deformity. Then, dull, aching thoracic back pain may start, which is typically located between the scapulae and is aggravated by physical activity, prolonged sitting, standing, and forward flexion. The severity of pain and progressive worsening of the kyphosis have poor correlation. Studies have also shown that patients with SD complain of back pain and pain of greater intensity more frequently than healthy individuals.^{58,62} Garrido et al.⁶⁴ identified that patients with SD most frequently reported thoracic and shoulder pain, followed by thoracolumbar and lower back pain. The prevalence of severe thoracic back pain has been reported by Bradford⁶⁵ in 50% of adult SD patients; however, Sorenson⁵¹ reported that thoracic pain that is seen in 50% of adolescent patients decreases to 25% in adults. So it may be assumed that the typical pain of SD tends to diminish as the adolescent approaches skeletal maturity. This also supports the psychosomatic aspects of pain in adolescence. As the aesthetic concerns tend to decrease after adolescence, the reported pain also decreases.

Excessive kyphosis usually results in nonstructural hyperlordosis of the lumbar spine, anterior PT, and protracted shoulders. Pain can be caused by a variety of factors. Scheuermann's lesions, such as disk damage or inflammatory lesions, can cause pain, especially in kyphotic deformities. LBP is usually caused by muscle tension, but it can also have an identifiable cause, like spondylolysis, as previously mentioned. The thoracolumbar form of the disease corresponds to a lowering of the apex of the kyphosis at the thoracolumbar junction (T11–T12) and loss of LL for the lumbar form.⁶⁶ In a computerized keyword review, Summers et al.⁶⁷ reported that as

much as 80% of the radiologically suggested/diagnosed patients showed features of lumbar Scheuermann's disease and only 20% suggested classical Scheuermann's disease.

Thoracolumbar and lumbar forms of SD can be more painful than the classical form. Cosmetic deformity may be negligible due to less vertebral wedging, but more prominent Schmorl's nodes are usually seen. Another important aspect of this subgroup is that the lower apex of the deformity tends to cause a loss of LL and may affect the sagittal spinopelvic balance more frequently than the classical type.⁶⁸ A cervical form of Scheuermann's disease has not been described, which might be explained by the fact that uncus, which is not mobile during puberty, could protect the vertebral endplate against mechanical stress.⁵⁵

Pain is often the most noticeable symptom for adult patients with SD that can last for months or even years. It is often associated with a set of symptoms such as tightness of the chest muscles, hamstrings, and hip flexors, and worsens after activity and at the end of the day. It has also been reported that multiple Schmorl's nodes are highly associated with lumbar disk disease and lower back pain.⁶⁹ Sward et al.⁷⁰ analyzed back pain and radiological changes of the thoracolumbar spine in 142 top Swedish athletes: they reported significant correlation between back pain and the number of different radiological changes as reduced disk height, Schmorl's nodes and change of configuration of vertebral bodies. These radiological abnormalities occurred in 36%–55% of the athletes in the thoracolumbar region. It is suggested that pain observed in the presence of Schmorl's nodes is related to (1) inflammatory changes induced by the presence of intraspongious disk components in contact with the vertebral bone marrow, and/or (2) trabecular bone fractures which may further aggravate inflammation and edema.⁷¹

Quality of life in SD

In the literature, patients with SD have been reported to experience more back pain and other back-related constraints than the normal population.⁶² In addition, these patients have been reported to experience cosmetic problems and psychological issues at a higher rate when compared with the normal population.⁷² Damborg et al.⁷³ found no statistically significant difference in perceived mental health status among SD patients. However, in perceived physical health status, they found a significant difference between SD patients and a control group. Patients with SD scored significantly worse on physical status.⁷³

Jonsson et al.⁷⁴ evaluated the prevalence of Scheuermann's disease, and their pain, in Swedish elderly men. In his study, among men with SD and back pain, none reported severe pain, 57% moderate, and 43% mild, compared with 7%, 50%, and 44% in those without SD. In addition, likewise, 63% patients with SD reported no sciatica, 15% sciatica without neurological deficits, and 22% sciatica with neurological deficits, compared with 56%,

16%, and 28% in those without the disease. These data clearly indicate that the condition is not associated with neck or back pain in the elderly population.

Nonsurgical treatment

Usually, the condition stops progressing once a child is older and has stopped growing. Recommended treatment options may depend on the severity of the spinal curve and the limitations on flexibility and mobility.⁷⁵ Generally, skeletally immature patients with a kyphotic curve less than 50° are candidates for physical therapy and bracing treatment. Intensive physiotherapy exercise programs for postural improvement have been tried for many years; however, no study has yet determined that physical therapy is sufficient on its own to decrease the severity of the kyphosis.

Bracing can be an effective treatment option for patients with moderate-to-severe kyphotic deformities. In adolescent patients with a kyphosis of <60°, exercise programs with or without part-time bracing are recommended. In patients with a kyphosis of >60°, a brace program of >20 h per day should be considered.⁷⁵ The goal of bracing is to prevent further progression of the curvature and to provide support for the spine. However, the specific type of brace and duration of use will depend on the individual patient and the degree of the curvature.

Lifestyle modifications are also considered as an important part of conservative treatment. The majority of teens with SD will find relief with some combination of lifestyle modifications and stretching exercises.⁷⁶ Sports associated with jumping, marked stress, and functional overuse of the back, especially in patients with thoracolumbar and lumbar Scheuermann's kyphosis, should be discouraged.⁷⁵

Surgical treatment

Most widely accepted indications for surgical intervention include progressive kyphosis despite brace treatment, neurologic deficit, persistent pain, or notable deformity in a skeletally mature individual. More clear indications for surgical intervention are still being investigated in the literature. Polly et al.⁷⁷ found that surgically treated patients were older and heavier. Children who underwent surgical treatment were less satisfied with their preoperative appearance and were more likely to be experiencing pain at the apex of the deformity. They observed that there was no detectable difference in maximal sagittal Cobb angle between operatively and conservatively treated groups. Overall, the authors concluded that maximal Cobb angle is not a notable predictor for surgical intervention, but pain and patient dissatisfaction played important roles in determining when to proceed with surgical intervention.⁷⁷ According to Koller et al.,⁷⁸ satisfactory clinical results can be obtained with both anteroposterior and posterior-only spinal fusion techniques (Figure 7); however, in

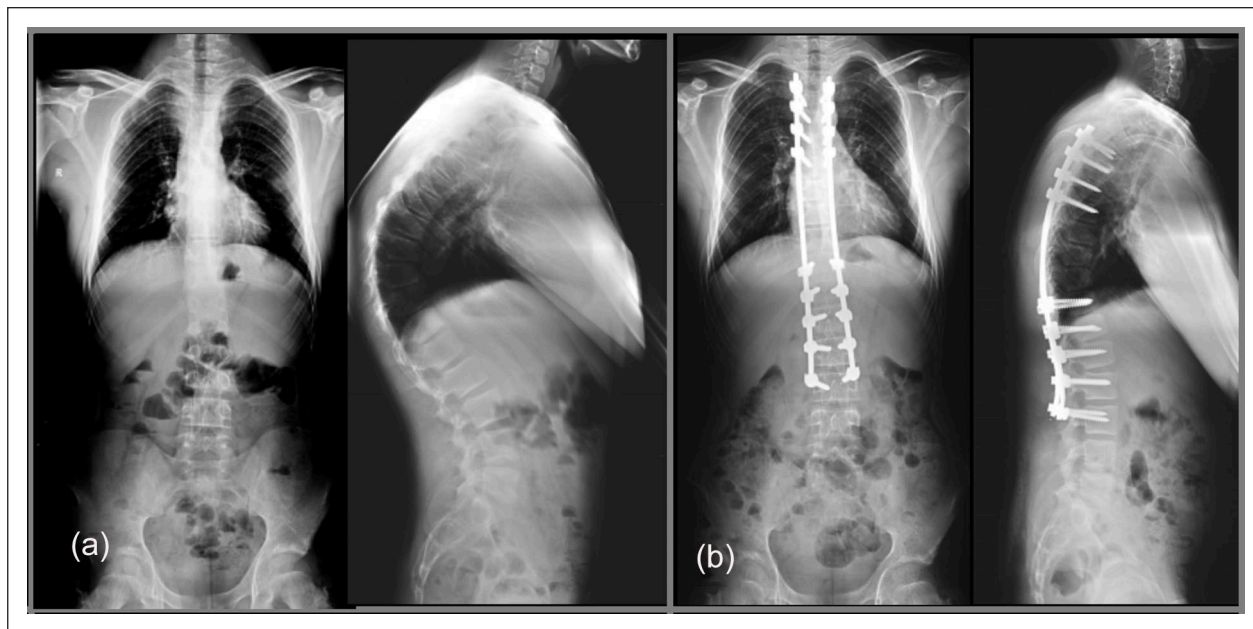


Figure 7. (a) A 15-year-old male patient with 85° of kyphosis and significant back pain. (b) Posterior instrumented fusion with multiple Ponte osteotomies was performed, reducing the kyphosis to 48°.

selected patients with severe deformity, posterior-based three-column osteotomies may be required to achieve adequate correction if the surgeon chooses a posterior-only approach (Figure 6).

The goal of surgical management is to correct the deformity and to improve function. It must be noted that a high incidence of postoperative complications related to alignment is observed, especially when compared with adolescent idiopathic scoliosis (AIS).⁷² The reported rates of proximal junctional kyphosis (PJK) and distal junctional kyphosis (DJK) are very high even in the best centers and range from 24.2% at the midterm follow-up to 53% at the long-term follow-up, frequently leading to unplanned surgeries.^{79,80} Selection of instrumentation levels is of utmost importance in the surgical treatment of SD, as emphasized by a recent study concluding that if the upper instrumented level is below T2, risk of PJK will increase.⁸¹ Lonner et al.⁸² compared health-related quality of life (HRQOL) measures and sagittal deformity in patients who were operatively treated for SD or AIS, and healthy controls. They reported that patients with SD demonstrated significantly decreased scores in all subdomains (function/activity, mental health, self-image/appearance, and satisfaction with management) of the SRS-22 questionnaire compared with patients with AIS and healthy controls.⁸² These studies all point out to the importance of avoiding unnecessary surgical interventions.

Summary—SD

SD is a disease of unknown etiology and is a common cause for pediatric back pain. Diagnosis is relatively

straightforward, and advanced imaging is rarely required. Similar to spondylolysis/spondylolisthesis, most of the patients are successfully managed with conservative measures alone. It must be kept in mind that unlike a defect in the pars, the kyphotic deformity in SD is not expected to resolve which may have social and functional implications during adulthood.

Indications for surgical management are not clearly defined in the literature. When surgery is considered, decision must be made carefully especially considering the fact that most adolescents' main complaints are pain and cosmesis, which may resolve in adulthood without surgery. Posterior-only approach is successfully utilized by most surgeons, but complications are not rare and it may be challenging to acquire and maintain a proper sagittal balance.

Association of spondylolysis/spondylolisthesis and SD

It is not very unusual for different spinal disorders to be seen concomitantly, which is also true for spondylolysis and Scheuermann's disease. This co-occurrence has been brought into attention by Ogilvie and Sherman,⁸³ who reported that in patients with Scheuermann's disease, spondylolysis was also seen at the lower lumbar levels in 50%. Increased LL seen in Scheuermann's disease may result in increased stresses on the pars region, but the true mechanism for this co-occurrence has not been revealed.

Both these disorders affect the sagittal plane, and if one is encountered by the physician, a thorough evaluation of the whole spine is necessary to look out for other problems.

Conclusion

Prevalence of LBP in childhood is proven to be much more frequent than it was once believed. Spondylolysis/spondylolisthesis and SD are two of the most frequent disorders causing back pain in this age group, even co-occurrence is possible. Another shared feature is that in their severe forms, both these disorders may disrupt the patient's sagittal alignment.

These disorders generally follow a benign course and conservative measures successfully manage the symptoms in most cases. However, when surgery is indicated, careful technical consideration is necessary to prevent unnecessary complications and acquire a well-balanced spine. Meticulous planning of the technical details is of utmost importance for a good outcome.

Author contributions

Riza Mert Cetik: Literature review and manuscript drafting.

Michal Latalski: Literature review and manuscript drafting.

Muharrem Yazici: Conception, supervision, critical review of the manuscript.

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