Is there a place for surgical management in adolescents with Scheuermann's disease?

F.X. Lambert, C. Decante, E. Mayrargue, S. Guillard, A. Chalopin, A. Hamel

Introduction

Scheuermann's disease, or osteochondritis deformans juvenilis dorsi, was first described in 1920 by Danish orthopedic surgeon and radiologist Dr Holger Wefel Scheuermann who reported a stiff thoracic hyperkyphosis in young apprentice watchmakers [1]. Scheuermann's disease is one of the most frequent causes of back pain in adolescents [2-6], with an incidence varying between 0.04 and 10% [2,7-10]. Nevertheless, an increase in prevalence was observed between 2003 and 2012, increasing from 3.6 to 7.5 per 100,000 [11]. Scheuermann's disease occurs most frequently in children aged 8 to 12 years old, with stiffer forms concerning primarily the ages 12-16 years [9]. There is a clear male predominance with a male to female ratio of 2:1 [11].

The etiology of Scheuermann's disease remains unknown. Mechanical factors have been considered with associations to a bad posture or obesity, but no evidence has been found. According to a Danish cohort study on 35,000 twins, 74% of cases were hereditary [12] and 1.8% were due to a syndrome, mostly Prader-Willi and Marfan's syndrome [11].

The treatment of Scheuermann's disease includes physiotherapy, orthopedic treatment with bracing, and occasionally surgery. Scheuermann's disease is considered a developmental disorder as it progresses during growth. Nevertheless, the natural history of this disease during adulthood remains unknown, thus making surgical intervention controversial.

Clinical features

Scheuermann's disease is characterized by thoracic or thoracolumbar hyperkyphosis associated with lumbar and cervical compensatory hyperlordosis in order to maintain proper sagittal balance. Progression toward stiffness may cause pain and important aesthetic complaints in adult patients. Hamstring and iliopsoas tightness, and stiffness of the shoulder girdle may also be associated. Furthermore, one third of patients present with non-structural scoliosis and/or L5-S1 spondylolisthesis [2].

There is often confusion between Scheuermann's disease and postural kyphosis. Clinically, patients with postural kyphosis have a flexible kyphotic deformity and do not present with hamstring tightness [13].

Psychological distress may also be associated with Scheuermann's disease and is often underestimated. In a series of 1,070 pediatric subjects, Hom et al. found an associated 5.5%

rate of depression and 4.5% rate of anxiety in patients with Scheuermann's disease, compared to only 1.7% and 0.8% in the general population, respectively [11].

Radiographic features

The radiographic diagnosis of Scheuermann's disease relies on certain criteria established by Sorensen in 1964 [14]. Anterior wedging across 3 consecutive vertebrae superior to 5° confirms the diagnosis [14]. Vertebral endplate irregularities with intervertebral disc herniations through the vertebral endplate (Schmorl nodes) may also be found [2,15].

The stiffness of the kyphotic deformity is best evaluated on hyperextension views.

Alterations in spinal sagittal balance on whole spine sagittal radiographs must be evaluated by drawing a vertical line passing through the center of the auditory meati. This line usually passes just posterior to the femoral heads. Global sagittal alignment is also assessed by measuring the spino-sacral angle, spinal tilt, and spino-pelvic tilt [16]. Alterations in spinal sagittal balance have a much more significant impact on the quality of life of these patients than on aesthetics and determine the progression of the disease [17-20].

MRI may be useful as a complement to conventional radiographs in order to assess the severity of inter-vertebral disc disease and to evaluate the entirety of the Schmorl nodes and endplate irregularities. Finally, spinal cord anomalies that may or may not be secondary to Scheuermann's disease can also be screened [5,21,22].

Natural history of Scheuermann's disease

Scheuermann's disease is a spinal pathology occurring primarily during the later stages of growth, with a natural history that remains difficult to elucidate. Nonetheless, symptoms tend to decrease during adulthood, whereas a slight increase in the kyphotic deformity may be observed during aging. However, in some patients, the kyphotic deformity progresses during adulthood and causes, besides important aesthetic consequences, mechanical pain and neurological complications [23].

Bartynski et al. studied the mean thoracic kyphosis and lumbar lordosis in patients without spinal disease [24]. Their results showed that, in young adults (18-35 years old), thoracic kyphosis was in average 27°, whereas in patients aged 65 years or older, it was 42°. Other authors have suggested that a "normal" kyphosis varies between 27 and 44° in adolescents [15,25], and between 20 and 50° in adults [26-28]. Stagnara et al. suggested that a "normal" kyphosis or lordosis does not exist, and that these values are only indicative and not normative [28].

In a study published in 2017, Ristolainen et al. followed 19 patients with Scheuermann's disease who were treated non-operatively with an average follow-up of 46 years and concluded that the deformity progressed slowly [29]. This progression, however, did not predict the appearance of symptoms. Mean age at final follow-up was 64.7 years, and patients had a mean kyphosis of 60°. In their series, kyphosis progressed by 14° but non-uniformly. Progression of the kyphotic deformity <10° was found in 42% of patients, and >20° in 32% of patients. A progression of 32° was reported in 3 cases (one female aged 65 years old in whom kyphosis increased from 48° to 80° over 36 years; two males, one aged 73 years old in whom kyphosis increased from 28° to 60° and one aged 76 years old in whom kyphosis increased from 50° to 82° over 59 years). The severity of the kyphotic deformity at the time of diagnosis did not predict progression of the deformity. No significant differences in terms of quality of life were found in patients with more progressive curves. Moreover, a significant increase in vertebral body wedging and lumbar lordosis was observed.

In a study conducted on 67 subjects with Scheuermann's disease and a control group, Murray et al. found that, after a mean follow-up of 32 years, patients who had not been operated presented certain functional restrictions but without major limitations to activities of daily living [30]. Patients with Scheuermann's disease also reported more back pain compared to controls, with no limitations in activities of daily living or during work. In fact, no differences were found based on the type of work, the number of days absent from work due to low-back pain, aesthetic complaints, painkiller use, participation in hobbies, and the presence of numbness in the lower extremities. Furthermore, patients with thoraco-lumbar deformities presented with more functional limitations than purely thoracic deformities. No differences were found between subjects with Scheuermann's disease and controls in terms of marital status, but patients with a kyphosis >85° were more frequently single and had a lower pulmonary capacity.

In a study conducted on 49 subjects with Scheuermann's disease who were treated non-operatively with a mean follow-up of 37 years, Ristolainene et al. found a higher prevalence of back pain and limitations in activities of daily living compared to the general population [31]. However, this increased prevalence was not correlated to the severity of the kyphosis. In fact, no differences in the intensity of the back pain or functional limitations were found between subjects with a kyphosis <40° and >60°.

Treatment

Conservative treatment

The treatment of Scheuermann's disease depends primarily on the intensity of the pain, the development of neurological or cardiopulmonary complications, aesthetic considerations, and the degree and progression of the deformity, all the while taking into account the residual growth of the spine.

Physiotherapy includes softening of the hips and stretching of the hamstrings as well as the spinal erectors and stabilizers. Although physiotherapy does not slow the progression of the disease, it is still recommended for symptomatic patients presenting with a stiff curve and may even be complementary to bracing in order to counteract the stiffness of the deformity. In a study conducted on 351 subjects with Scheuermann's disease aged 17 to 21 years old, Weiss et al. found a significant decrease of pain (16-32%) with physiotherapy, thereby suggesting the positive effects of this type of management on the primary complaint of this disease in young adults.

Treatment by bracing in patients during growth may improve the kyphotic deformity and even lead to vertebral remodeling. Nonetheless, once the brace has been weaned, a loss of correction is often observed and may even reach 30% [30-34]. Flexible kyphosis, early management with a deformity <65°, an initial correction >15° with a brace, and a residual growth of the spine of at least 1 year are factors of good prognosis when treatment with a brace is considered [32,33]. A stiff kyphosis with a deformity >65°, vertebral wedging >10°, and a finished spinal growth are considered as risk factors for failure of treatment by bracing.

Surgical treatment

Indications

Indications for surgical management are controversial, and objective assessments are scarce in the literature. The large variability of the natural history of Scheuermann's disease renders an estimation of the risk-benefit ratio of surgical management difficult to establish. Additionally, surgery in Scheuermann's disease is complex and may lead to potentially serious complications [34]. According to the Scoliosis Research Society, less than 1% of spine surgeries are conducted on patients with Scheuermann's kyphosis [35]. The frequency of patients with Scheuermann's disease who are managed operatively is actually stable [11], with recently published series indicating an apparent increase [36].

Although the severity of the deformity in the sagittal plane is a major criterion leading to surgical management in patients with Scheuermann's disease, the angular kyphotic value does not seem to be the primary factor in surgical decision-making. The threshold of the sagittal Cobb angle indicating the need for surgery varies greatly between publications [34,37-44]. Furthermore, Polly et al. found no differences in maximal Cobb angle between operated (70°) and non-operated (73°) subjects [45]. The majority of authors recommend surgical management in progressive deformities that are superior to 60-75° and not controlled by bracing, in patients with back pain resistant to lifestyle modifications (physical activity, NSAIDS >6 months), appearance of neurological or cardiopulmonary complications, or in case of significant aesthetic complaints [23,34,38-40,46-55]. Patients and their families must be made aware of the expected benefits and the risks of surgery. The evaluation of global sagittal alignment is paramount, especially in thoraco-lumbar deformities, since these locations disrupt the harmony of the different curves in the sagittal plane and alter the sagittal alignment of the spine.

Care must be taken as parents of children with Scheuermann's disease suffering from back pain and aesthetic complications may pressure surgeons into adopting certain treatment modalities [45].

Surgical technique: Anterior, posterior or combined approach?

In 1975, Bradford et al. were the first to report a series of 22 patients who were treated surgically by posterior fusion using only the Harrington technique [56]. They reported a mean correction of 25° (reduction from 72 to 47°) and a mean loss of correction of 21° in 16 patients (72% of cases) [23].

In order to avoid this loss of correction, a combined approach with anterior release and posterior fusion was long considered the gold standard in treatment and was particularly recommended for the treatment of severe and stiff deformities [37,43,46,55,57-61]. As other surgical options were developed, specifically pedicle screws, very satisfying results by posterior approach only have been reported. In fact, multiple studies have concluded that the quality of the correction is comparable between posterior-only and combined approaches.

In fact, combined approaches are associated with higher complication rates than a posterior-only approach [7,11,41,62]. The posterior-only approach leads to less blood loss, decreased operative times, and seems to decrease the risk of adjacent segment disease [42,62,63]. In their case series, Riouallon et al. found no significant differences in complication rates between the two methods and reported 3 types of complications that were specific to the anterior approach [61]. Mizashahi et al. did not show any complications in patients treated with a posterior-only approach [64]. Furthermore, the length of hospital stay was longer for patients operated by a combined approach [11,36].

The majority of authors report no significant loss of correction in the posterior-only approach compared to the combined approach [58,61,62,65,66], except for Temponi et al. who found that the combined approach showed less loss of correction [7]. As a result, the number of patients with Scheuermann's kyphosis treated by posterior fusion without anterior release has significantly increased from 34 to 78% between the years 2000 and 2008 [11,36]. Furthermore, complication rates have decreased from 22.6% to 15.5% during the same time frame (a rate that still remains especially high) [11,36].

Ponte or Smith-Peterson osteotomies during a posterior-only correction allows for better correction [57,58,64,65,67]. Correction of the deformity by posterior compression without osteotomy lengthens the anterior column of the spine and increases the risk of spinal cord elongation and of anterior spinal artery spasms. One or more osteotomies around the apex of the deformity would shorten the posterior column without lengthening the spine anteriorly, and thereby decrease the risk of neurological complications.

Implant density has also been evaluated. It is commonly believed that an increase in the density of the implants favors the correction of kyphosis. However, Behrbalk et al. showed a similar correction between an implant density of 100% and 50% around the apex of the deformity [38]. The decrease in implant density is associated with a decrease of complications by 50% and surgical cost by 32% [38]. Finally, no significant differences were found between a posterior-only approach and a combined approach in terms of pain, functional outcome, and aesthetic satisfaction [61,62].

Level of instrumentation

Surgery for Scheuermann's disease must include the entire kyphotic deformity. The choice of proximal and distal instrumentation levels and the degree of correction is not clearly elucidated in the literature. For some authors, correction of the kyphotic deformity must not surpass 50% [34], with a post-operative thoracic kyphosis ranging between 40 and 50° [16,43,53,68-70]. In fact, over-correction increases the risk of proximal junctional kyphosis (PJK), whereas under-correction would maintain the compensatory hyperlordosis at the lumbar level which would accelerate spinal degeneration in the long term [71,72]. In order to prevent the occurrence of PJK, the upper instrumented vertebra should be chosen as the most proximal vertebra that is included within the thoracic kyphotic deformity [55,60].

Recently, some authors have studied the relationship between pelvic and sagittal parameters. An analysis of these parameters allows to predict the estimated post-operative lumbar lordosis, and by conjunction, the thoracic kyphosis [16,19,69,70,73]. It has been shown that patients with Scheuermann's disease developing PJK after correction of their hyperkyphosis were those with a high pelvic incidence and a significant post-operative deficit in lumbar lordosis [41,74]. As a result, the correction of thoracic kyphosis must take into account the pelvic incidence. Nasto et al. have established an equation for the prevention of such a phenomenon [74]: %LL correction = 0.66 x (%TK correction) – 2.

The choice of lower instrumented vertebra remains controversial in the literature. This choice must preserve the maximum of lumbar mobility, all the while preventing distal adjacent segment disease, especially distal junctional kyphosis (DJK). DJK has not been well-defined in the literature. Zhu et al. defined DJK as a sagittal Cobb angle >10° between the superior endplate of the lower instrumented vertebra and the inferior endplate of the immediately adjacent distal vertebra [75]. Another risk factor for the development of DJK is a significant shift from lordosis to neutral or kyphosis of the intervertebral disc immediately distal to the lower instrumented vertebra. In a meta-analysis by Gong Y et al. evaluating four studies and 173 patients, 20.8% of patients who were operated for Scheuermann's disease developed DJK post-operatively, and 27.8% of those with DJK required revision surgery [76].

Cho et al. developed the concept of the stable sagittal vertebra (SSV) [77]. This is defined as the most proximal lumbar vertebral body bisected by the vertical line from the posterior-superior corner of the sacrum. The first lordotic vertebra (FLV) is defined as the vertebra lying immediately distal to the most proximal lordotic disc. SSV and FLV can sometimes be superimposed. On the one hand, the choice of the FLV as the lower instrumented vertebra does not lead to higher incidences of DJK and allows for the preservation of distal motion segments [15,53,78]. On the other hand, the choice of the SSV as lower instrumented vertebra decreases the risk of adjacent segment disease [76,77,79-82]. In fact, in a series including 20 subjects with Scheuermann's disease operated by spinal fusion ending on the FLV, Cobden et al. found that 15% of patients developed DJK postoperatively [65].

In subjects with Scheuermann's disease with a thoraco-lumbar kyphotic curve, Zhu et al. found that ending the construct on the FLV was sufficient, but that in thoracic curves, it was necessary to include the SSV in order to decrease the risk of DJK [75].

Complications

Complication rates after surgery for Scheuermann's disease are especially high. Around 10% of subjects treated by posterior fusion report at least one complication, and 20% of those treated by a combined approach [11,36]. Huq et al. published a meta-analysis including 1,829 subjects with Scheuermann's disease treated by surgery between the years 1950 and 2017 [66]. Correction of the kyphotic deformity was associated with significant neurological complications up to 8% in this meta-analysis. The authors reported 25% instrument failure, 14% PJK, and 14% DJK in posterior-only approaches, with 10% requiring revision surgery, as well as 26% PJK, 20% DJK, 21% respiratory complications, and 6% cardiovascular complications in combined approaches, with 11% requiring revision surgery.

Conclusion

Scheuermann's disease is a deformity of the growing spine with a relatively benign long-term natural history. Nevertheless, the deformity may progress in adulthood and lead to mechanical pain and neurological complications such as adult spinal deformity.

It is difficult to predict which patients will be symptomatic during adulthood, and which patients could benefit from surgical treatment. A comprehensive analysis of the sagittal alignment must be undertaken before proposing operative treatment, which presents with a high rate of severe complications. If surgery is considered, recent data suggest the need for vertebral osteotomies and posterior spinal fusion ending on the stable sagittal vertebra.

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