ELSEVIER

The Spine Journal ■■ (2016) ■■–■■

THE SPINE JOURNAL

Case Report

Growing rod erosion through the lamina causing spinal cord compression in an 8-year-old girl with early-onset scoliosis

Fahad H. Abduljabbar, MBBS, FRCSC[a,b], Feras Waly, MBBS[a,c], Anas Nooh, MBBS[a,b], Jean Ouellet, MD, FRCSC[a,*]

[a]*Division of Orthopedic Surgery, McGill Scoliosis and Spine Centre, McGill University Health Centre, 1650 Cedar Ave, T8-200, Montreal, QC H3G 1A4, Canada*

[b]*Department of Orthopedic Surgery, King Abdulaziz University, Abdullah Sulayman St, P.O. Box 80215, Jeddah, 21589, Saudi Arabia*

[c]*Division of Orthopedic Surgery, University of Tabuk, Khalid Elsedery St, P.O. Box 741, Tabuk, 71491, Saudi Arabia*

Received 3 December 2015; revised 29 March 2016; accepted 4 May 2016

Abstract

BACKGROUND CONTEXT: Early-onset scoliosis often occurs by the age of 5 years and is attributed to many structural abnormalities. Syndromic early-onset scoliosis is considered one of the most aggressive types of early-onset scoliosis. Treatment starts with serial casting and bracing, but eventually most of these patients undergo growth-sparing procedures, such as a single growing rod, dual growing rods, or a vertical expandable titanium prosthetic rib.

PURPOSE: This case report aimed to describe an unusual complication of erosion of a growing rod through the lamina that caused spinal cord compression in an 8-year-old girl with early-onset scoliosis.

STUDY DESIGN: This is a case report.

METHODS: A retrospective chart review was used to describe the clinical course and radiographic findings of this case after rod erosion into the spinal canal.

RESULTS: The patient underwent successful revision surgery removing the rod without neurologic complications.

CONCLUSIONS: Patients with syndromic early-onset scoliosis are more prone to progressive curves and severe rotational deformity. We believe that the severe kyphotic deformity in addition to the dysplastic nature of the deformity in this population may predispose them to this unusual complication.

Keywords: Complications; Early-onset scoliosis; Growing spine; Growth sparing spinal instrumentation; Growing rods; Implant migration; Rod migration; Scoliosis

Introduction

Early-onset scoliosis (EOS) has been used to describe the presence of scoliosis of all etiologies by the age of 5 years [1–3]. The causes of EOS include neuromuscular conditions, congenital vertebral anomalies, associated syndromes, and structural lesions of the central nervous system [1]. Early-onset idiopathic scoliosis is a diagnosis of exclusion when these patients have no structural abnormalities. The initial treatment of EOS starts with serial casting, followed by bracing based on the principle of prevention of curve progression [4–8]. Despite such treatment, however, these curves may progress, necessitating surgical intervention. The disadvantages of serial casting and bracing include compromise of chest wall development and subsequent worsening of cardiopulmonary function [9,10]. Treating children with progressive EOS is challenging in many aspects [11]. The remaining

FDA device/drug status: Not applicable.

Author disclosures: ***FHA:*** Nothing to disclose. ***FW:*** Nothing to disclose. ***AN:*** Nothing to disclose. ***JO:*** Consulting: DePuy Synthes (B, Paid to the author), outside the submitted work; Speaking and/or Teaching Arrangements: AO Foundation (B, Paid to the author), outside the submitted work; Endowments: Synthes (F, Paid to the institution), outside the submitted work; Grants: Shriners International (E, Paid to the institution), outside the submitted work; Fellowship Support: AO Foundation (E, Paid to the institution), outside the submitted work.

The disclosure key can be found on the Table of Contents and at www.TheSpineJournalOnline.com.

No funds were received in support of this work. No benefits in any form have been or will be received from a commercial party related directly or indirectly to the subject of this article. FA (None). FW (None). AN (None). JO (grants from AO Foundation, DePuy Synthes; personal fees from DePuy Synthes, outside the submitted work).

* Corresponding author. Shriners Hospital for Children-Canada, McGill University, 1003 Décarie Boulevard, Montreal, Canada H4A 0A9. Tel.: +001 514 412 4464; fax: +001 514 412 4353.

E-mail address: jean.ouellet@muhc.mcgill.ca (J. Ouellet)

http://dx.doi.org/10.1016/j.spinee.2016.05.001

potential for growth makes it harder to fuse their spines to control the curve progression without losing that potential. This can compromise lung development, which will result in respiratory insufficiency. Thus, focus on EOS spine surgery has shifted from the spine alone to concomitant consideration of the chest wall and lungs, and the goals of treatment include a well-aligned spine and a thoracic cavity sufficiently developed to support adequate pulmonary development and function [12].

After this recognition of the importance of pulmonary function, the pendulum shifted toward preservation of spine and chest growth through the use of growth-sparing techniques. These techniques rely on the principle of a distraction-based construct to control curve progression, allowing axial and chest growth simultaneously. Many techniques have been described, such as the use of a single growing rod [13,14], dual growing rods [14], and a vertical expandable titanium prosthetic rib [15,16].

The growing rod technique is associated with a high rate of complications [17–19].

These include rod breakage, hook displacement, wound infections, and to a lesser extent neurologic injury [11]. We report an unusual case of an 8-year-old girl who underwent a dual growth lengthening for EOS that was complicated by erosion of a growing rod into the spinal canal.

Case report

The patient was an 8-year-old girl who initially presented to our clinic at the age of 25 months as a case of EOS. She was diagnosed with camptomelic dwarfism, Pierre Robin syndrome, severe kyphoscoliosis (Fig. 1), and spina bifida occulta. Bracing was initially started but failed to control progression of the curve. Casting was next attempted but also failed to prevent progression. Thus, at the age of 2 years and 10 months, the patient underwent halo gravity traction for 4 months. At the time of halo application, proximal and distal

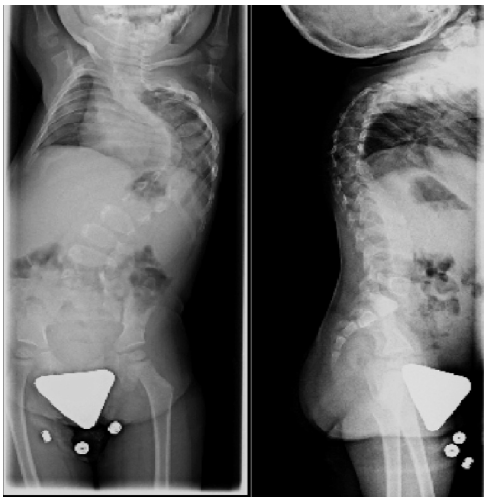

Fig. 1. Posteroanterior (PA) and lateral views of the whole spine showing severe kyphoscoliosis.

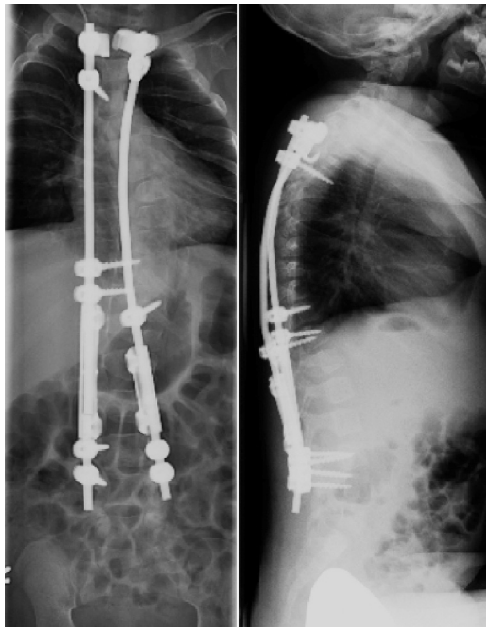

Fig. 2. Posteroanterior (PA) and lateral views of the spine showing posterior spinal fusion (PSF) of two-level focal sites, T1–T3 and L3–L4, and growing rods.

spinal anchors were inserted for selective fusions of two levels, T1–T3 and L3–L4. These were done at this time, as patient had no posterior elements at the distal segments and severe kyphosis, wanting to have solid fixation before the placements of the growing rods. Once partial correction was achieved by halo traction, growing rods were inserted with additional gliding anchors at the apex [5] to maximally translate and correct the spinal deformity (Fig. 2). At the age of 3 years and 7 months, the patient started to develop an iatrogenic low-grade slip at the level of L5–S1, which remained asymptomatic. She underwent growing rod lengthening every 6 months until she ran out of length at the age of 5 years, at which time she underwent a revision of her growing rods. At the age of 7 years, the L5–S1 spondylolisthesis has progressed to a high grade (Fig. 3), and the patient started to complain of right-sided L5 radiculopathy. Advanced imaging, including computed tomography (CT) scanning and magnetic resonance imaging (MRI), revealed high-grade spondylolisthesis and right L5 nerve root compression. Surprisingly, the imaging also showed that the left rod was eroding through the lamina at the apex of her thoracic deformity and encroaching on the spinal cord (Fig. 4). Despite this complication, the patient had no signs of myelopathy. At that point, a decision was made to extract the left rod, perform a thoracic spinal decompression and right L5 nerve root decompression, and extend the fusion level distally to the pelvis.

Under a total intravenous anesthetic, the patient was positioned prone on the Jackson table. Even before initiating surgery, there were no motor evoked potentials. An emergency wake-up test was done and the child did not move her

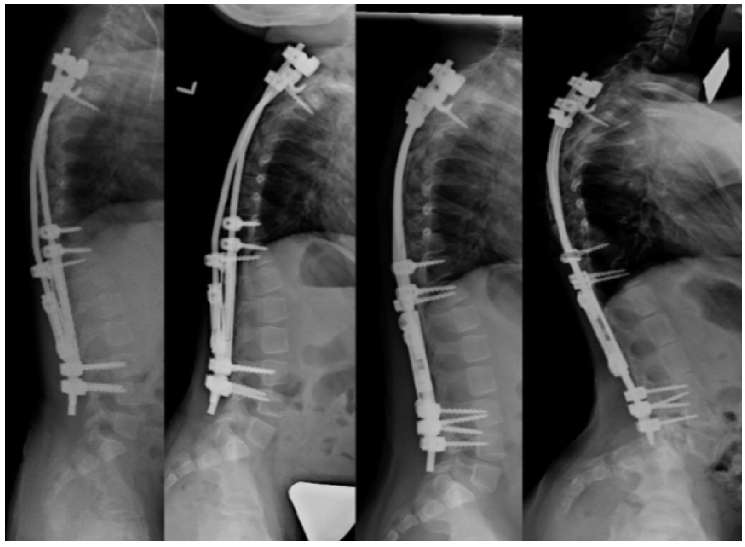

Fig. 3. Serial lateral views over 5 years showing the progression of L5–S1 spondylolisthesis from a low-grade to a high-grade spondylolisthesis.

legs, and hence the procedure was aborted. In recovery, the patient was moving both lower extremities. A week later, with maintained high mean arterial pressure, we returned to surgery and we extracted the left rod, performed a thoracic spinal decompression and right L5 nerve root decompression, and extended the fusion level distally to the pelvis.

A rod-to-rod connecter was used to place the left rod away from the midline so as not to site on top of the posterior elements and the spinal canal. At that point, lengthening was attempted, but all waves on the motor evoked potentials were lost, and the lengthening procedure was aborted. The patient was awoken and her neurologic exam revealed some long-tract signs; however, she had normal strength in both lower extremities. The patient's gait was slightly abnormal preoperatively, which we believed to be secondary to spondyloptosis. Her gait returned to normal postoperatively. The power of the lower extremity was normal and symmetrical. At the patient's last follow-up at the age of 8 years and 8 months, the results of her neurologic exam were normal and the spine construct appeared stable (Fig. 5).

Discussion

Treatment of patients with EOS is often challenging. Skeletally immature patients with significant scoliotic curves and the high potential for progression mandate intervention to stop the progression. Unfortunately, the conventional posterior spinal fusion technique results in reduced cardiopulmonary function. As a result, distraction-based posterior spinal instrumentation has been advocated to correct scoliosis and allow for spinal growth and thoracic development [20]. Bilateral growing rods are effective in EOS, stabilizing spinal

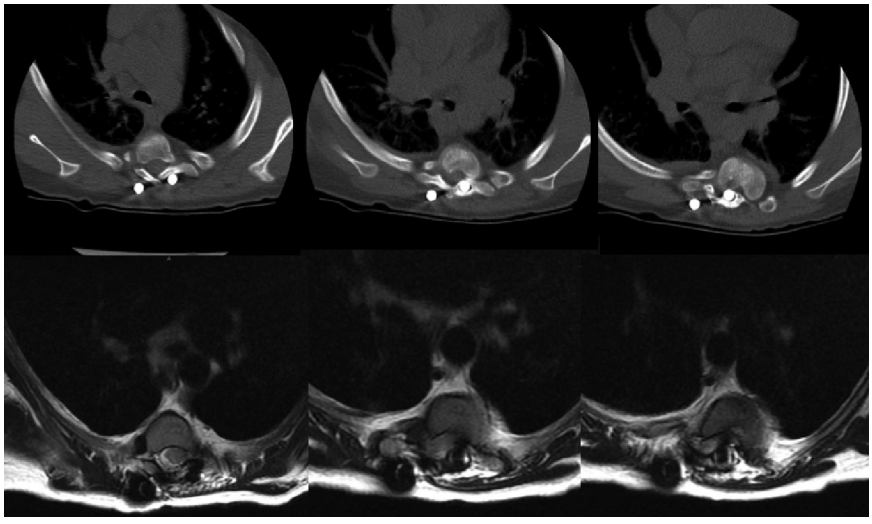

Fig. 4. Axial computed tomography (CT) scan cuts and magnetic resonance imaging (MRI) T2-weighted images demonstrating the left rod eroding through the lamina at the apex of the thoracic deformity and encroaching on the spinal cord.

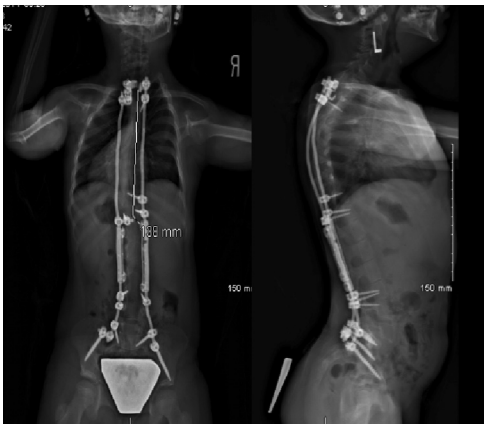

Fig. 5. Final x-rays of the patient at the age of 8 years and 8 months showing a stable construct and the rod-to-rod connector on the left side.

deformity while achieving height gain close to that of physiological growth [21]. Vertical expandable prosthetic titanium rib can also be used to manage EOS but are better suited for patients with chest malformation or thoracic insufficiency syndrome [13,14,22]. Considering our patient had normal chest walls, we opted for a spine-based growing rod construct. It has been shown that in either growth-sparing techniques, repetitive surgeries increase the risk of overall complications rate [23]. These complications can be divided into six categories: wound complications, implant-related complications, alignment complications, fusion complications, neurologic complications, and medical or surgical complications. Wound complications may be either superficial or deep infection. Implant-related complications might involve dislodgement of hooks or screws or rod breakage. Prominent hardware can also lead to wound complications. Alignment complications include junctional kyphosis or curve decompensation. Fusion complications include unwanted fusion of adjacent levels or pseudoarthrosis of the foundation levels. The frequency of lengthening procedures depends on patients' growth. We do measure height and arm span to quantify their overall growth in every visit, then do their lengthening accordingly [24,25]. If the patient is going through a growth spurt, then we tend to do the lengthening in less than 6 months to catch up with the growth spurt and allow distraction for spine growth subsequently. Age and sex will not affect decisions on frequency of imaging or interventions. As a general rule, they should not exceed 6 months if gain in height is to approximate physiological growth (1.2–1.7 cm per year) [13].

This case report is the first of its kind in the management of EOS. It describes bone remodeling of the posterior spine elements leading to a growing rod migration through the lamina, which in turn led to canal encroachment and subsequent spinal cord compression. Surprisingly, the patient did not show any signs of spinal cord compression. This finding was completely incidental on the advanced imaging during the assessment of an iatrogenic L5–S1 spondyloptosis. Spinal deformities in patients with campomelic dysplasia are usually progressive [11]. They also have either absence or delayed ossification of the thoracic pedicles [12]. We hypothesize that the significant kyphoscoliotic curve and her dysplastic posterior elements may have predisposed her to this complication.

Rod migration into the spinal canal is an unusual complication; however, it has been described previously in the literature. Four case reports describe this complication, but in patients with very different underlying diagnoses: one patient with an L1 fracture dislocation [26], two adult patients with treated idiopathic scoliosis [27,28], and one patient with neuromuscular scoliosis [29]. The rod migration into the spinal canal in these cases was attributed to either biological (infection) or mechanical factors (kyphotic deformity, posterior elements defect, or micro-motion of the rod). In the two cases of adult idiopathic scoliosis [27,28], the authors concluded that progressive laminar erosion accompanied by extensive laminar remodeling explained the migration coupled with an underlying low virulence infection. They also attribute the migration of the rod into the canal because of the compressive forces exerted by the implant on the apex of deformity. Although the concurrent infection may have contributed to the laminar erosion, it is of particular note that the erosion was found only on the right side of the thoracic spine (side of convexity), whereas signs of infection were noted bilaterally on hardware removal. The concave rod remained well fixed, with no evidence of laminar erosion. In the other two cases, rod migration was related to mechanical reasons. The rod migration in the third cases was secondary to a laminar defect post a decompressive laminectomy for an L1 fracture dislocation [26]. In the fourth case, posterior spinal fusion was done to a skeletally immature cerebral palsy patient, and the authors attributed the rod migration to the patient's significant potential for growth. Using a spanning type of fixation allowed the thoracic spine to keep growing, and the presence of a relatively rigid construct enabled progressive migration of the rods toward the spinal canal, and the rotational forces acting in conjunction with compressive forces generated by the convex rod on the posterior arch most likely contributed to progressive migration and subsequent rod entrapment in the spinal canal [30]. Moreover, their hypothesis is that the baclofen pump in a patient with chronic muscular insufficiency and lack of muscular compensatory function further enhanced the pressure exerted by the two rods on the laminae, ultimately leading to pushing the rods to migrate toward the spinal canal. Three out of four cases had neurologic deficit on presentation, which warranted advanced imaging to diagnose such a complication [26,27,29]. Luckily, our patient did not suffer any neurologic deficits despite the significant spinal cord compression. The fourth case presented with signs of deep infection, which was evaluated by advanced imaging that confirmed the rod erosion into the spinal canal [28]. The delay to diagnose this complication in the reported cases ranged from 5 to 10 years, which reflects a slow process of posterior elements remodeling due to either mechanical or biological causes. Our case was diagnosed with this complication incidentally after 4 years of her first surgery.

Children who initially receive growing rod implants at a younger age are likely to undergo more procedures than children initially treated at an older age [21]. Bess et al. reported a 13% decrease in the likelihood of experiencing a complication for each year of increase in age at the initial procedure [20]. Unfortunately, we had to intervene surgically in this patient at the age of 3 years because the bracing and casting had failed to prevent curve progression. Fortunately, we did not encounter any wound complications.

The unusual complication reported here raises the question of the need for frequent assessment of growing rods by use of CT scanning or MRI for this patient population. There are no clear indications for advanced imaging unless patients are showing symptoms of neurologic compromise. We believe that in the face of EOS with underlying diagnosis of bony dysplasia and severe kyphosis, it may be warranted to get a cross-sectional imaging, such as a CT scan or an MRI, over the gibbous at 3- to 4-year intervals to avoid the delay in the diagnosis of implant migration and spinal cord compression. Such recommendation must not be taken lightly and needs to be discussed with the parents as using CT scans for children will put them at a greater risk for cancer than adults. From a standard given dose of radiation, children are at higher risk of developing cancer than adults. Children are inherently more radiosensitive and because they have more remaining years of life, during which a radiation-induced cancer could develop [31]. This cancer risk estimate for current CT use ranges from 1.5% to 2.0% [32]. This risk is higher in younger female patients [33]. The effort to avoid unnecessary risk of cancers must be multifaceted by avoiding unnecessary CT scans and eliminating the 13-fold difference in radiation dose for the same CT scan [34].

Conclusions

Patients with EOS require multiple growth-sparing procedures. Dysplastic conditions in these patients, associated with severe curves and kyphosis, may predispose to the rod encroaching on the spinal canal and subsequent spinal cord compression. Early recognition of this complication requires the identification of high-risk patients and follow-up with cross-sectional imaging if needed to avoid advanced spinal cord damage.

References

[1] Gillingham BL, Fan RA, Akbarnia BA. Early onset idiopathic scoliosis. J Am Acad Orthop Surg 2006;14:101–12.

[2] Dickson RA. Conservative treatment for idiopathic scoliosis. J Bone Joint Surg Br 1985;67:176–81.

[3] Dickson RA, Archer IA. Surgical treatment of late-onset idiopathic thoracic scoliosis. The Leeds procedure. J Bone Joint Surg Br 1987;69:709–14.

[4] Akbarnia BA. Management themes in early onset scoliosis. J Bone Joint Surg Am 2007;89(Suppl. 1):42–54.

[5] Ouellet J. Surgical technique: modern Luque trolley, a self-growing rod technique. Clin Orthop Relat Res 2011;469:1356–67.

[6] D'Astous JL, Sanders JO. Casting and traction treatment methods for scoliosis. Orthop Clin North Am 2007;38:477–84, v.

[7] McMaster MJ, Macnicol MF. The management of progressive infantile idiopathic scoliosis. J Bone Joint Surg Br 1979;61:36–42.

[8] Mehta MH. Growth as a corrective force in the early treatment of progressive infantile scoliosis. J Bone Joint Surg Br 2005;87:1237–47.

[9] Koumbourlis AC. Scoliosis and the respiratory system. Paediatr Respir Rev 2006;7:152–60.

[10] Sevastikoglou JA, Linderholm H, Lindgren U. Effect of the Milwaukee brace on vital and ventilatory capacity of scoliotic patients. Acta Orthop Scand 1976;47:540–5.

[11] Coscia MF, Bassett GS, Bowen JR, Ogilvie JW, Winter RB, Simonton SC. Spinal abnormalities in camptomelic dysplasia. J Pediatr Orthop 1989;9:6–14.

[12] Khoshhal K, Letts RM. Orthopaedic manifestations of campomelic dysplasia. Clin Orthop Relat Res 2002;401:65–74.

[13] Campbell RM Jr, Smith MD, Hell-Vocke AK. Expansion thoracoplasty: the surgical technique of opening-wedge thoracostomy. Surgical technique. J Bone Joint Surg Am 2004;86-A(Suppl. 1):51–64.

[14] Emans JB, Caubet JF, Ordonez CL, Lee EY, Ciarlo M. The treatment of spine and chest wall deformities with fused ribs by expansion thoracostomy and insertion of vertical expandable prosthetic titanium rib: growth of thoracic spine and improvement of lung volumes. Spine 2005;30(17 Suppl.):S58–68.

[15] Campbell RM Jr. Operative strategies for thoracic insufficiency syndrome by vertical expandable prosthetic titanium rib expansion thoracoplasty. Oper Tech Orthop 2005;15:315–25.

[16] Hell AK, Campbell RM, Hefti F. The vertical expandable prosthetic titanium rib implant for the treatment of thoracic insufficiency syndrome associated with congenital and neuromuscular scoliosis in young children. J Pediatr Orthop B 2005;14:287–93.

[17] Klemme WR, Denis F, Winter RB, Lonstein JW, Koop SE. Spinal instrumentation without fusion for progressive scoliosis in young children. J Pediatr Orthop 1997;17:734–42.

[18] Mineiro J, Weinstein SL. Subcutaneous rodding for progressive spinal curvatures: early results. J Pediatr Orthop 2002;22:290–5.

[19] Blakemore LC, Scoles PV, Poe-Kochert C, Thompson GH. Submuscular Isola rod with or without limited apical fusion in the management of severe spinal deformities in young children: preliminary report. Spine 2001;26:2044–8.

[20] Bess S, Akbarnia BA, Thompson GH, et al. Complications of growing-rod treatment for early-onset scoliosis: analysis of one hundred and forty patients. J Bone Joint Surg Am 2010;92:2533–43.

[21] Odent T, Ilharreborde B, Miladi L, et al. Fusionless surgery in early-onset scoliosis. Orthop Traumatol Surg Res 2015;101(6 Suppl.):S281–8.

[22] Samdani AF, St Hilaire T, Emans JB, et al. The usefulness of VEPTR in the older child with complex spine and chest deformity. Clin Orthop Relat Res 2010;468:700–4.

[23] Akbarnia BA, Emans JB. Complications of growth-sparing surgery in early onset scoliosis. Spine 2010;35:2193–204.

[24] Luk KD, Saw LB, Grozman S, Cheung KM, Samartzis D. Assessment of skeletal maturity in scoliosis patients to determine clinical management: a new classification scheme using distal radius and ulna radiographs. Spine J 2014;14:315–25.

[25] Sanders JO, Browne RH, Cooney TE, Finegold DN, McConnell SJ, Margraf SA. Correlates of the peak height velocity in girls with idiopathic scoliosis. Spine 2006;31:2289–95.

[26] Quint DJ, Salton G. Migration of Luque rods through a laminectomy defect causing spinal cord compression. AJNR Am J Neuroradiol 1993;14:395–8.

[27] Obeid I, Vital JM, Aurouer N, et al. Intraspinal canal rod migration causing late-onset paraparesis 8 years after scoliosis surgery. Eur Spine J 2014. doi:10.1007/s00586-014-3367-y.

[28] Tribus CB, Garvey KE. Full-thickness thoracic laminar erosion after posterior spinal fusion associated with late-presenting infection. Spine 2003;28:E194–7.

[29] Canavese F, Dmitriev P, Deslandes J, et al. Rod migration into the spinal canal after posterior instrumented fusion causing late-onset neurological symptoms. J Pediatr Orthop 2015 [e-pub ahead of print].

[30] Hell AK, Campbell RM, Hefti F. [New treatment concept for children with thoracic insufficiency syndrome due to congenital spine deformity]. Klin Padiatr 2005;217:268–73.

[31] Brenner DJ, Hall EJ. Computed tomography—an increasing source of radiation exposure. N Engl J Med 2007;357:2277–84.

[32] Berrington de Gonzalez A, Mahesh M, Kim KP, et al. Projected cancer risks from computed tomographic scans performed in the United States in 2007. Arch Intern Med 2009;169:2071–7.

[33] Smith-Bindman R, Lipson J, Marcus R, et al. Radiation dose associated with common computed tomography examinations and the associated lifetime attributable risk of cancer. Arch Intern Med 2009;169:2078–86.

[34] Redberg RF. Cancer risks and radiation exposure from computed tomographic scans: how can we be sure that the benefits outweigh the risks? Arch Intern Med 2009;169:2049–50.