Spine

SPINE Volume 46, Number 24, pp 1696–1704

Minimally Invasive Surgery for Neuromuscular Scoliosis: Results and Complications at a Minimal Follow-up of 5 Years

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Study Design. A prospective study.

Objective. The aim of this study was to report the results of an alternative technique to growing rods (GR) for neuromuscular scoliosis using a minimally invasive fusionless surgery with a minimum of 5 years' follow-up.

Summary of Background Data. Conservative treatment is not effective in progressive neuromuscular scoliosis. Early surgery using GR is increasingly advocated to control the deformity while preserving spinal and thoracic growth before arthrodesis. These techniques still provide a high rate of complications.

Methods. The technique relies on a bilateral double rod sliding instrumentation anchored proximally by four hooks claws and distally to the pelvis by iliosacral screws through a minimally invasive approach. The clinical and radiological outcomes of 100 consecutive patients with neuromuscular scoliosis who underwent this fusionless surgery with a minimum follow-up of 5 years were reviewed.

Results. 6.5 ± 0.7 years after initial surgery, six patients were lost of follow-up and 11 died of unrelated raison. Of the 83 remaining patients at latest follow-up, mean Cobb angle was stable to 35.0° which correspond to 61% correction of the initial deformation. Mean pelvic obliquity was 29.6° (0.3°–80.0°) preoperatively and 7.2 (0.2°–23.5°) at latest follow-up. Correction of the hyper kyphosis remained stable. Skeletal maturity was reached in 42 of 83 patients (50.6%). None of these patients has required spinal fusion. The global complication rate was 31.3%.

Conclusion. The outcomes of this minimally invasive fusionless technique at 5 years follow-up showed a stable correction of spinal deformities and pelvic obliquity over time, with a reduced rate of complication. The arthrodesis was not required for all patients at skeletal maturity. This technique could be a good alternative to arthrodesis for neuromuscular scoliosis.

Key words: bipolar technique, fusionless surgery, growth guided surgical technique, minimally invasive fusionless surgery, neuromuscular scoliosis.

Level of Evidence: 3

Spine 2021;46:1696–1704

Neuromuscular scoliosis is caused by a disorder of the brain, spinal cord, or muscular system. Neuromuscular curves are often associated with trunk imbalance, pelvic obliquity, and kyphosis, starting to develop at an early age and usually worsen over time despite conservative treatment.[1] Common conditions associated with neuromuscular scoliosis are cerebral palsy, myelomeningocele, spinal muscular atrophy, and muscular dystrophy. Early posterior spine fusion (PSF) used to be performed for the most severe of them, with the risk of stunting trunk growth and concomitant effects on lung development. Arthrodesis provides a high complication rate, especially infections and hemorrhages requiring long intensive care unit stay.[2,3] Growth-sparing surgical procedures have therefore been developed to preserve spinal and thoracic growth and to postpone arthrodesis.[4,5] These techniques were first introduced by Harrington in 1962.[6] They became more and more popular during the two last decades with the advent of traditional growing rods (TGR). After initial surgery, some surgical procedures of rod lengthening are performed to achieve as spinal height as possible, followed by a PSF when the patient reaches skeletal maturity.[7,8] However, the complication rate can reach 40% to 73%[9–12] with these methods.

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Acknowledgment date: November 22, 2020. First revision date: January 9, 2021. Second revision date: February 21, 2021. Acceptance date: March 23, 2021.

The device(s)/drug(s) is/are FDA-approved or approved by corresponding national agency for this indication.

No funds were received in support of this work.

Relevant financial activities outside the submitted work: grants.

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DOI: 10.1097/BRS.0000000000004082

An original technique of bipolar instrumentation was developed as an alternative to TGR for the treatment of neuromuscular scoliosis.[13] The surgical approach was minimally invasive, since proximal and distal short incisions were performed thus preserving the intermediate area to avoid early fibrosis and autofusion. The results were maintained after 2-year follow-up with a 61% correction of the Cobb angle and 83% correction of the pelvic obliquity. The aim of this work was to evaluate the clinical and radiological outcomes of minimally invasive fusionless technique for neuromuscular scoliosis at a minimum of 5 years of follow-up.

PATIENTS AND METHODS

Study Design and Data Collection

Medical charts of the first 100 consecutive patients who underwent surgery for neuromuscular scoliosis at our department between 2011 and 2015 were reviewed. The study was approved by the appropriate ethics committee (CPP, ID-EUDRACT #2014-A01043–44). Informed consent was obtained from parents/guardians of all participants. The original minimally invasive bipolar technique used in these patients has been previously described.

Patients were operated under intraoperative traction and evoked potential monitoring. Two short incisions, one thoracic and one lumbar, were made to insert the anchors. The instrumentation extends from T1 to the sacrum, proximally with a double pediculosupralaminar claw and distally why iliosacral screws. Two long precurved rods (diameter 5.5 mm) are inserted in an intramuscular way from the proximal wound to the distal one and attached proximally to the hooks and distally to two short rods fixed inside the iliosacral connectors. Side-to-side connectors and cross-links connect the rods to build a solid sliding frame construct.

In this technique, the correction is obtained through distraction maneuvers that are applied at the distal part of the sliding construct, without further rod manipulations. The bipolar concept relies on a continuous tension between the proximal and the distal points of fixation obtained with rod lengthening on demand. Rods were lengthened using the previous distal incision to access the side-to-side connectors. Lengthening procedure was performed according to the following indications:

- to perfect pelvic correction in case of major residual pelvic obliquity, in particular thanks to the possibility of performing asymmetrical lengthening of the rods;
- to improve a persistent trunk imbalance in the frontal or sagittal plane.

The amount of rod lengthening was froml5 to 30 mm.

For each patient, the following radiographic parameters were recorded before surgery, immediately after surgery, and at last follow-up: stages of skeletal maturity as reflected by the Risser sign, Cobb angle, pelvic obliquity, and thoracic kyphosis. Screw loosening was defined as a radiolucent area (≥1 mm in circumference) around the screw, noted on plain radiograph by at least two observers. Radiographic measurements were performed using previously described methods.[14,15] All types of complications were reviewed including implant failure (*e.g.,* malposition of the ilio-sacral screw, hooks migration, and implant breakage) and infections. Mean body weight was recorded preoperatively and at latest follow-up.

Statistical Analysis

Variables were described as mean (range). Changes in radiological parameter values from baseline to last follow-up were evaluated by applying the pairwise Student t test. P values <0.05 were considered significant. Complications were described as n (%).

RESULTS

Demography

Of the first 100 patients, six were lost of follow-up and 11 died of independent causes from spinal surgery. As such, 83 patients (83% of the initial number of patients) were included (40 females, 43 males). All patients were Risser stage 0 at initial surgery with a mean age of 11.5 years (5.2–15.4). The diagnoses were cerebral palsy (n = 45), spinal muscular atrophy (n = 21), muscular dystrophy (n = 10), and other neuromuscular disorders (n = 7). Mean follow-up was 6.5 years (5.3–9.0). Thirteen patients were ambulatory. Mean body weight was 24.4 ± 6 kg preoperatively and 37.3 ± 7 kg at latest follow-up. (Table 1)

The average operative time for the initial surgery was 2 hours 26 minutes (from 1 hour 33 minutes to 3 hours 39 minutes; the average operative time for rod lengthening was 44 minutes (from 26 minutes to 1 hour 10 minutes.

Twenty-four percent of patients required one to two blood transfusions at initial surgery, none after rod lengthening.

Radiological Outcomes

Tables 2 and 3 report the changes in these radiological parameters.

Seventeen patients did not require rod lengthening thanks to a stable and satisfying correction after initial surgery. The other patients underwent one rod lengthening (30), two rod lengthening (24), three rod lengthening (11), or maximum four rod lengthening (1) to achieve progressive correction of spinal deformity and/or pelvic obliquity.

Screw loosening was visible radiographically in 17 patients (seven ambulatory and 10 nonambulatory) (Figure 1) No clinical or mechanical consequences of osteolysis were seen in any patients, and no revision was needed.

At last follow-up, skeletal maturity (Risser stage 4) was reached in 42 of 83patients (50.6%) with a mean of 2.9 ± 1.4 years after initial surgery. Mean Cobb angle was 34.5° (min 6.4° to max 51.7°) and mean pelvic obliquity was 7.4° (min 0° to max 20.3°). At last follow-up, none of these

TABLE 1. Main Features of the 100 Patients Treated With Fusionless Surgery for Neuromuscular Scoliosis

Loss of follow-up, n	6/100
Unrelated death	11/100
Follow-up duration, y, mean (range)	6.5 (5.3–9.0)
Age at surgery, y, mean (range)	11.5 (5.2–15.4)
Sex (F/M)	40/43
Diagnosis, n	
Cerebral palsy	45
Spinal muscular atrophy	21
Muscular dystrophy	10
Other neurological conditions	7
Nonambulatory/ambulatory, n	70/13
Body weight, kg	
Preoperatively	24.4 ± 6
At latest follow-up	37.3 ± 7
Rod lengthening per patients, mean	1.3 (0–4)
Interval between initial surgery and 1st lengthening, y, mean	1.6 ± 0.6
Interval between lengthenings, y, mean	1.3 ± 0.6
Interval between final lengthening and initial surgery, y, mean	2.8 ± 1.0

TABLE 2. Radiographic Data of the 83 Patients

	Preoperative	Postoperative	Last Follow-up	*P*
Cobb angle, °, mean (range)	89.0° (46.1°–149.4°)	33.0° (5.0°–65.8°)	35.0° (6.4°–68.5°)	0.33
Pelvic obliquity, °, mean (range)	29.6° (0.3°–80.0°)	12.2° (0.1°–35.2°)	7.2° (0.2°–23.5°)	0.28
Hyper kyphosis T4-T12 >50° mean (range)	69.3° (51.8°–102.1°)	32.0° (13.1°–52.1°)	31.3° (8.8°–50°)	0.31
T1-S1 length, mean, cm	30.02 (22.30–40.13)		36.0 (29.3–44.2)	

TABLE 3. Radiographic Data of Patients at Skeletal Maturity

Patients at skeletal maturity (Risser >4), n, %	42/83 (50.6%)
Interval between initial surgery and skeletal maturity, y, mean	2.9 ± 1.4
Interval between skeletal maturity and latest follow-up, y, mean	3.7 ± 1.1
Cobb angle, °, mean (range) at last follow-up	33.9° (6.4°–53.4°)
Pelvic obliquity, °, mean (range) at last follow-up	6.9° (0–19.9°)
Hyper kyphosis T4-T12 >50° mean (range) at last follow-up	23.2° (14.3°–37.2°)

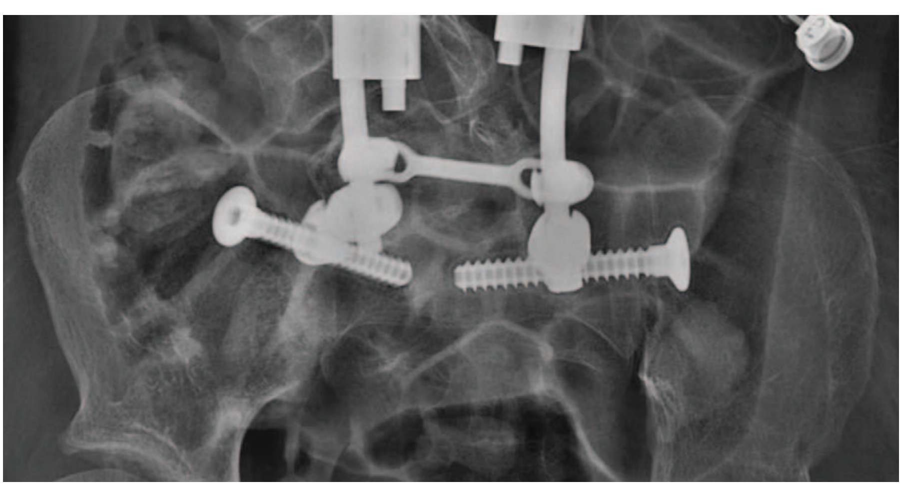

Figure 1. Screw loosening around iliosacral screw.

TABLE 4. Complications With Unplanned Surgery of the Minimally Invasive Surgery for Neuromuscular Scoliosis (n = 27, in 26/83 Patients)

Mechanical complications (n = 12)	
Proximal hook dislodgment	3
Malposition of iliosacral screw	4
Rod breakage	5
Cerebral palsy	
Ambulatory/ nonambulatory status	5/5
Mean body weight	1/4
	43.3 ± 6 kg
Infectious complications (n = 15)	
Diagnosis	
Cerebral palsy	11
Spinal muscular atrophy	3
Muscular dystrophy	1
Mean body weight, kg	
Preoperative	21.7 ± 4 (*P* > *0.05)*
Last follow-up	35.1 ± 6 (*P* > *0.05)*
Age at initial surgery	10.7 ± 2 (*P* > *0.05)*
Delay	
Within the first 3 mo of the initial surgery	3
After rod lengthening procedures	12
Management	
Irrigation and debridement, with appropriate antibiotic therapy	11
Implant removal followed by a new instrumentation	2
Implant removal without new instrumentation	2

patients has required PSF with a mean interval of 3.7 ± 1 year after skeletal maturity.

Complications

Twenty-seven complications (12 mechanical and 15 infections) occurred in 26 of 83 patients corresponding to a global rate of 31%. Table 4 details the 12 mechanical complications and 15 infections requiring unplanned surgery.

Mechanical Complications

Mechanical complications included three cases of proximal hook migration, four cases related to the malposition of iliosacral screw, and five cases of rod breakages.

Rod breakages occurred in a mean interval of 4.0 ± 1 year after initial surgery and were located at lumbar area. Of these patients, pattern curves were thoracolumbar left (three), thoracolumbar right (one), and double major (one) with no significant difference compared with the rest of the cohort ($P > 0.05$). All patients suffered from cerebral palsy with severe dystonia or hyperactivity. Mean body weight of these five patients was 43.3 ± 6 kg ($P = 0.04$) and one of them was a walking patient.

Infectious Complications

Of the 15 wound infections, three occurred within the first 3 months of the initial surgery and 12 after rod lengthening procedures. Eleven were successfully treated by irrigation and debridement, with appropriate antibiotic therapy. Four patients required implant removal because of chronic infection: two of them necessitated a new instrumentation 1 year later because of a deformity worsening; the two others did not need new implantation thanks to a stable correction of the deformity. There was significant difference related to mean body weight and age at initial surgery compared with the other patients of the cohort. Most of patients (11/15) suffered from cerebral palsy with a significant difference ($P = 0.039$).

DISCUSSION

In this study, the outcomes and complications of bipolar minimally invasive surgery in children with neuromuscular scoliosis were reviewed with a mean follow-up of 6.5 years. Results showed stable correction of spinal deformity and pelvic obliquity over time, including patients at skeletal maturity. PSF was not required for any patient at last follow-up.

This technique was initially used as an alternative to TGR to reduce mechanical and infectious complications while waiting for PSF planned at skeletal maturity for all of them. However, stable clinical and radiological evolution were noticed with a reduced rate of mechanical complications that did not require performing PSF as initially planned while obtaining an acceptable growth of the spine. Moreover, fibrosis and spontaneous fusion around the implants and along the rods was observed during revision surgeries

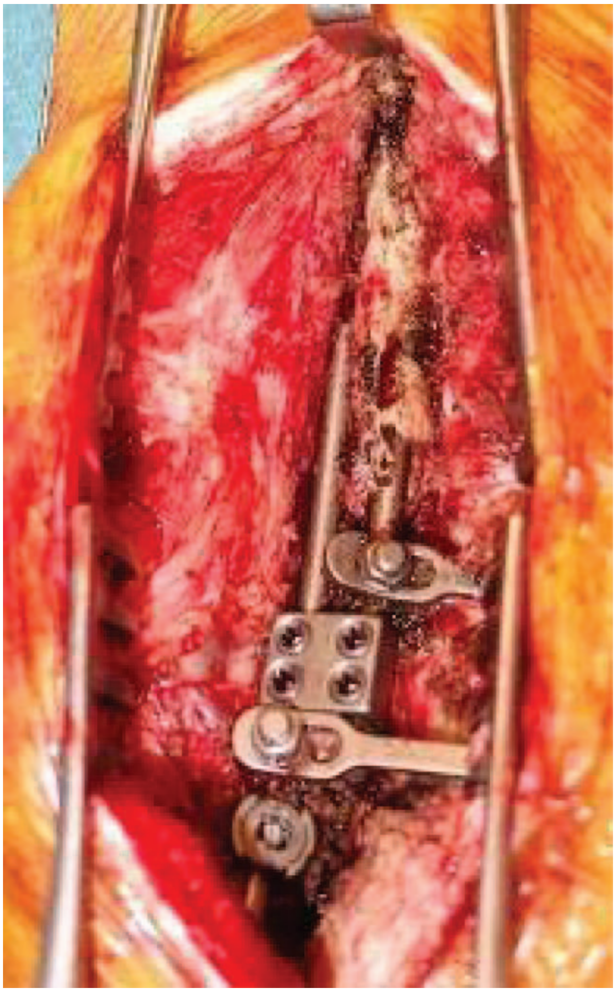

Figure 2. Fibrosis and spontaneous fusion observed around the implants during a rod lengthening procedure.

Figure 3. Computed tomography scan after 5 years' follow-up with autofusion around the rods.

for rod lengthening (Figure 2) and on complementary examinations such as computed tomography (CT) scan (Figure 3) or x-rays.

This delayed spontaneous fusion was asserted by lateral bending x-rays performed after hardware removal because of chronic infection, in one patient 8 years after initial surgery. The dynamic x-rays showed the total stiffness of the spine completely fused (Figure 4A–D). These observations lead to hypothesize that a progressive stiffening of the spine occurs over time under the influence of the permanent presence of rigid metallic rods (Figure 3). However, this concept of autofusion is not new. In 1984, Moe *et al*[16] noted spontaneous fusion at the curve extremities and/or at the curve apex in young children treated for severe deformities with subcutaneous Harrington rod without fusion. Mardjetko *et al*[17] observed extensive fibrosis and laminar spontaneous fusions under the instrumented regions with Luqué rods, frequently involving the entire thoracic spine but also extending across the thoracolumbar spine. In Cahill *et al*[18] series, the rate of autofusion in children treated with growing rods (GR) was 89%. This autofusion was characterized by solid sheet of bone similar to a mature fusion mass and lead to only moderate correction at the time of definitive fusion. Jain *et al*[19] also noticed a progressive ankylosis after GR surgeries and suggested to avoid PSF at skeletal maturity for patients with satisfactory final alignment and trunk height, and no clinical or radiographic evidence of implant-related problems. Recently, Bouthors *et al*[20] considered retaining dual GR instead of PSF in patients at skeletal maturity with satisfactory deformity correction.

The autofusion in traditional GR occurs at early stage due to the damage of the intermediate area secondary to repetitive rod lengthening procedures. This autofusion is unwanted and participates to the "law of diminishing return."[21] That is why rod-lengthening procedure was recommended with an interval of 6 months. The PSF realized at the end of traditional GR series with large autofusion was required because the deformity correction was unsatisfactory. In these cases, PSF were frequently performed earlier, before skeletal maturity.

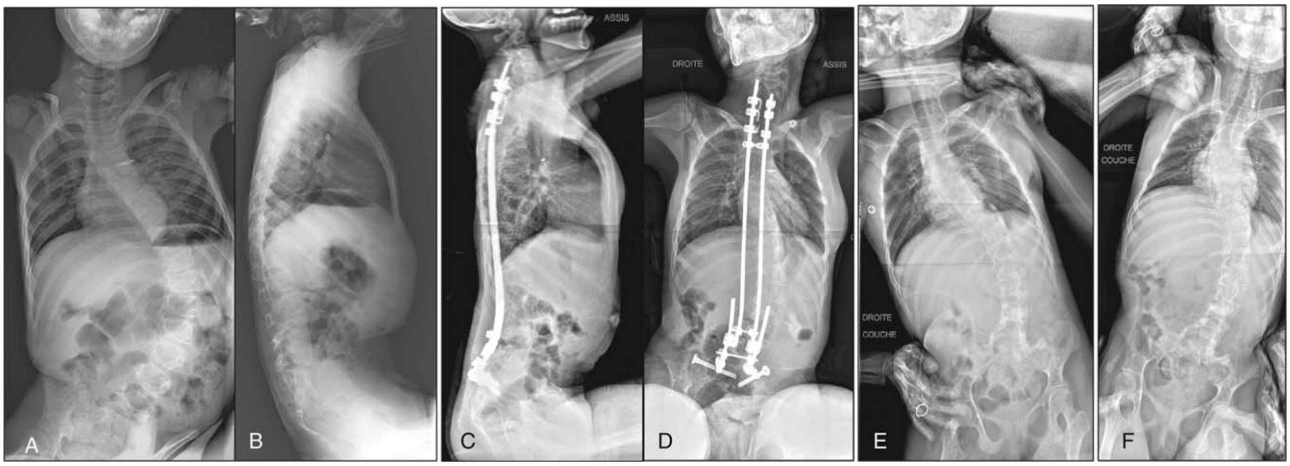

Figure 4. A 15-year-old girl with cerebral palsy. (**A** and **B**) Initial AP and lateral x-rays; (**C** and **D**) after initial surgery; (**E** and **F**) lateral bending after removal of instrumentation for chronic infection, 8 years after initial surgery.

In the bipolar minimally invasive technique, the autofusion is expected. It occurs at a delayed stage thanks to the preservation of the intermediate area between the two bipolar fixations and also thanks to the permanent presence of metallic rods. This phenomenon was experienced with Luqué-Trolley technique in the 70 s in which rods were sliding despite spontaneous fusion.[22,23]

The avoidance of PSF is particularly beneficial in this fragile neuromuscular population characterized by medical comorbidities, impaired bone quality, and poor nutritional status. Compared with other types of scoliosis, complication rates of PSF procedures in neuromuscular scoliosis show the highest rate of complications (24 to 75%), followed by congenital (10.6%) and idiopathic (6.3%) scoliosis.[24] More specifically, Connie Poe-Kochert *et al*[25] reported a mean of 1.5 complications per patient after PSF following treatment with the use of GR.

Unlike others GR techniques that are mainly provisory to wait for arthrodesis at skeletal maturity, this bipolar technique may be considered as an alternative to arthrodesis as it is strong enough to be definitive. The avoidance of PSF in our series relies on two main advantages of the instrumentation.

The first one is the relatively large deformity corrections of the Cobb angle (61%) and the pelvic obliquity (83%) which remained stable at latest follow-up (Figures 5A–D and 6). These amounts of correction were similar or better to TGR series[25,26] and even PSF.[27–31]

The second one is the strength and the stability of this type of fixation over time allowing PSF avoidance with a reduced mechanical complication rate. The bipolar construct differs from TGR by the stability of the proximal and distal fixations, the frame sliding construct, and the absence of intermediate area damage. Sponseller *et al*[9]

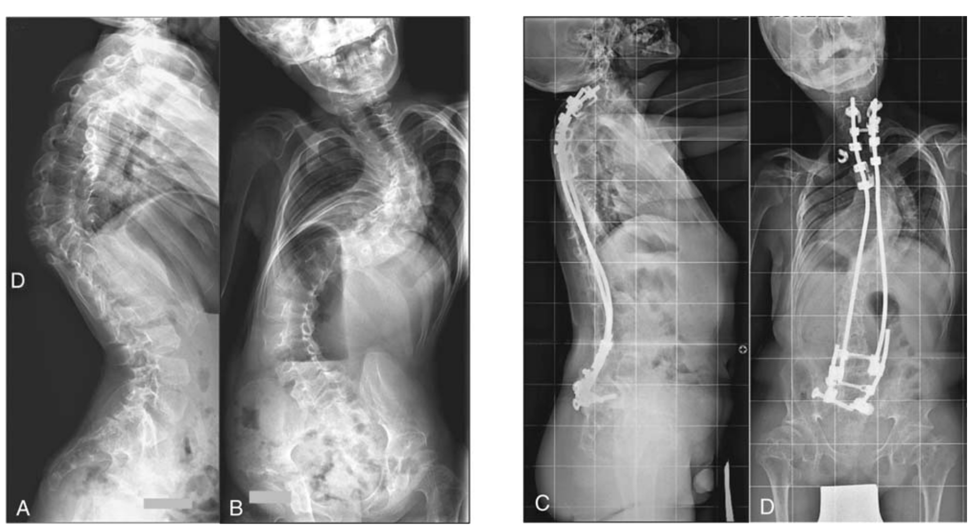

Figure 5. (A–D) Radiographs of a boy with cerebral palsy operated at 7 years' old. (**A** and **B**) Preoperative x-rays. (**C** and **D**) x-rays after 8 years' follow-up.

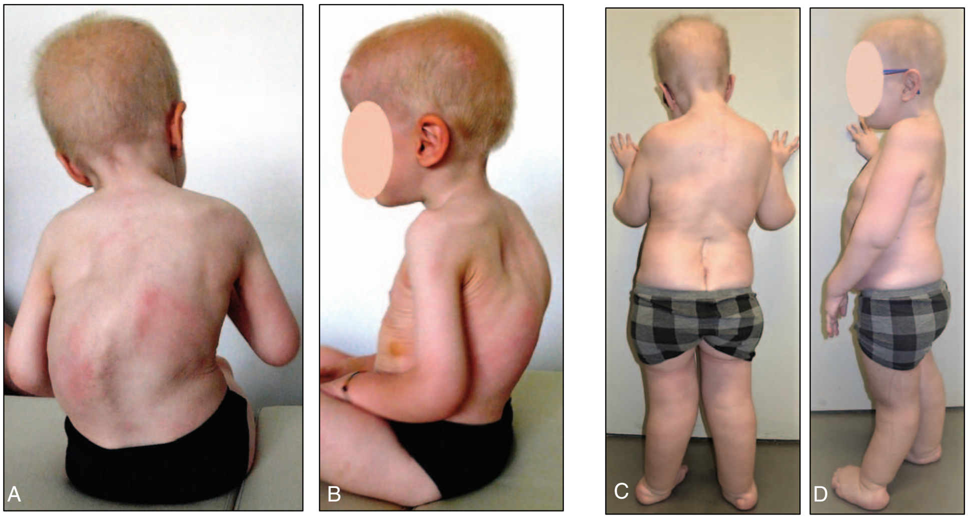

Figure 6. (A–D) Pre- and latest photographs after 8 years follow-up of the patient treated by the minimally invasive fusionless surgery.

reported a rate of 16.6% rod breakages in their series of 36 neuromuscular/syndromic patients treated with TGR extended to the pelvis. A rod breakage rate of 15% to 42% in TGR was published in early onset scoliosis from all etiology without pelvic fixation.[32–34] 10.6% were related to magnetically controlled GR.[10] In Mc Elroy series of GR for the treatment of scoliosis in cerebral palsy,[4] mechanical complications occurred in 19 of 27 (70%) patients including rod exchange (six), rod fracture (five), anchor revision (four), or anchor dislodgement (two).

In the present series, despite the absence of PSF, rod breakage occurred in five patients at risk (cerebral palsy with dystonia or hyperactivity, ambulatory patients and high body weight). They were managed by simple rod replacement or the use of four rods construct in the lumbar area as shown in Figure 7A–D. For one of them, it was decided peroperatively not to replace the broken rod because of the observation of extensive auto fusion around the rod. The removal of the rod could have generated important damages. At 3 years after the rod breakage, the spinal deformity of this patient remained stable without any complaint. Three cases of proximal hook dislodgement were experienced, one due to a severe osteoporosis and the others because of a lack of rod contouring at the beginning of our learning curve. This low rate of proximal mechanical complications can be explained biomechani-cally by the stability of the double claw-hook construct.[35] We experienced nine cases of proximal junctional kyphosis (11%) in our series without the need for revision surgery. This low incidence compared to other series[36] can be explained first by the type of the proximal fixation made of a solid double hooks claws; secondly by an adequate proximal bending of the rods; and finally by the high level of proximal limit of fixation (C7 or T1) in poor head control, sagittal trunk imbalance, and hyperkyphotic cases.

The construct also draws its strength and stability from its pelvic fixation technique using iliosacral screws, which have proved their efficiency and reliability for >40 years of experience.[29,37] No screw pullout or breakage was reported. Osteolysis around iliosacral screws was observed

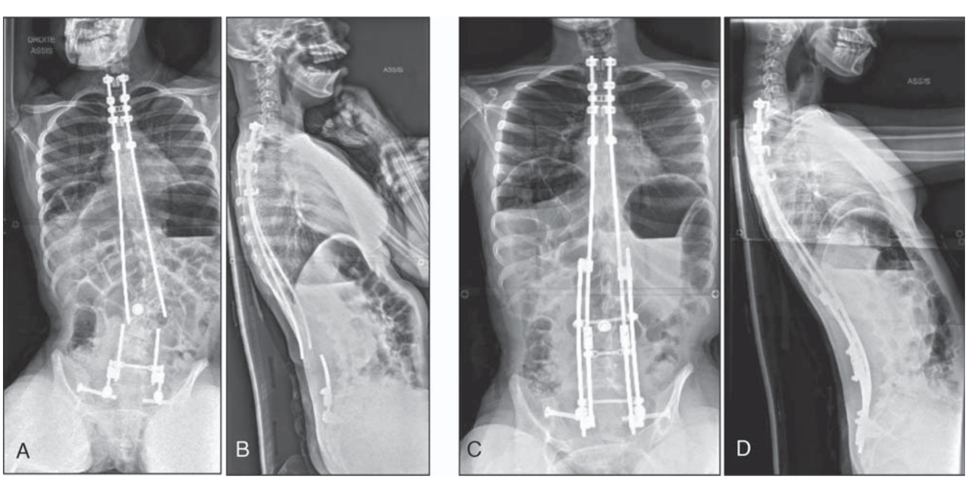

Figure 7. Nonambulatory 14-year-old girl suffering from dystonic cerebral palsy. (**A/B**) x-ray of rod fracture 4.5 years after initial surgery; (**C/D**) x-ray after revision surgery and additional rod implantation.

radiologically in 25 patients but no screw migration, pull-out, or adverse clinical outcomes was associated. The screw loosening was due to the microrotation of the screw concomitant to flexion/extension motion of the trunk of the patient. Iliosacral screw malposition was reported in four cases operated with freehand technique. No new case of misplaced screw was reported since the use of peroperative CT scan control.

Finally, the infectious rate of 18% was lower than TGR treatment in patients with cerebral palsy (30%)[4] or spinal muscular atrophy,[38] probably thanks to the minimally invasive approach preserving the intermediate area. The number of repeated surgical procedures is known as a high-risk factor especially in this population.[39] Three of 15 infections occurred after initial surgery and 12 after rod-lengthening procedures. This rate of infections after rod lengthening procedures could be reduced using self-expanding devices that avoid repeated surgeries.

One of the weakness of this study was the absence of comparative treatment to assess the lower rate of complications of the present surgical technique. Further work is under way to analyze long-term lung function and quality of life. Preliminary results suggest minimal impact of surgery on pulmonary management, and an overall patient satisfaction. All patients still did not achieve their skeletal maturity and the study still needs long-term follow-up (>10 years) to evaluate the effectiveness of this treatment to confirm the stability of the construct allowing the avoidance of PSF. Secondly, there were no radiological evidence to quantitatively assess the quality of the progressive ankylosis of the spine. These results open the way for further studies to evaluate spinal ankylosis using noninvasive examinations (*e.g.*, magnetic resonance imaging and ultrasound).

CONCLUSION

The outcomes of the bipolar minimally invasive fusionless technique in this series with a mean of 6.5 years' follow-up showed a stable and satisfying correction of spinal deformities and pelvic obliquity over time. The rigorous implantation of proximal and distal fixations contributed to reduce the rate of mechanical complications. In this series, half patients reached skeletal maturity and did not require PSF initially planned, leading to consider this technique as a viable alternative to PSF for neuromuscular scoliosis. However, longer follow-up is necessary to confirm these findings.

➢ Key Points

- ❑ Minimally invasive fusionless technique showed stable correction of spinal deformities and pelvic obliquity with a mean of 6.5 years' follow-up.
- ❑ A global complication rate of 31%.
- ❑ Patients at skeletal maturity did not require final posterior spinal fusion.
- ❑ This technique seems to be viable alternative to final posterior spinal fusion for neuromuscular scoliosis.

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