Original article

Fusionless surgery in early-onset scoliosis

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Abstract

Background: Surgical treatment of early-onset scoliosis has greatly developed in recent years. Early-onset scoliosis covers a variety of etiologies (idiopathic, neurologic, dystrophic, malformative, etc.) with onset before the age of 5 years. Progression and severity threaten respiratory development and may result in respiratory failure in adulthood. Many surgical techniques have been developed in recent years, aiming to protect spinal and thoracic development.

Material and methods: Present techniques are based on one of two main principles. The first consists in posterior distraction of the spine in its concavity (single growing rod, or vertical expandable prosthetic titanium rib [VEPTR]), or on either side (dual rod); this requires iterative surgery, for lengthening, unless motorized using energy provided by a magnetic system. The second option is to use spinal growth force to lengthen the assembly; these techniques (Luque Trolley, Shilla), using a sliding assembly, are known as growth guidance.

Results: These techniques are effective in controlling early scoliotic deformity, and to some extent restore spinal growth. However, they show a high rate of complications: infection, rod breakage, spinal fixation pull out and, above all, progressive spinal stiffness, reducing long-term efficacy. Respiratory gain is harder to assess, as thoracic expansion does not systematically improve respiratory function, particularly due to impaired compliance of the thoracic cage.

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1. Introduction

Surgical treatment of early-onset scoliosis has greatly developed in recent years. Early-onset scoliosis covers a variety of etiologies (idiopathic, neurologic, dystrophic, malformative, etc.) with onset before the age of 5 years. Progression and severity threaten respiratory development and may result in respiratory failure in adulthood. Due to severity of deformity and/or resistance to non-operative management, fusionless surgery may be performed to postpone arthrodesis, which, if performed early, prevents vertebral and thoracic growth. Many such techniques have been developed recently.

Present techniques are based on one of two main principles. The first consists in posterior distraction of the spine in its concavity (single growing rod [1], or vertical expandable prosthetic titanium rib [VEPTR] [2]), or on either side (dual rod) [3]; this requires iterative surgery, for lengthening, unless motorized using energy provided by a magnetic system. The second option is to use
spinal growth force to lengthen the assembly; these techniques (Luque Trolley [4], Shilla [5]), using a sliding assembly, are known as growth guidance.

Experience with these techniques, combined with fundamental research [6–8], has brought progress in the understanding of spinal and thoracic growth and pulmonary development, leading to a specific classification of these deformities [9] (Fig. 1).

The topic of the 2014 round-table of the Scoliosis Study Group (Groupe d’étude de la scoliose: GES) in Nice was “Fusionless surgery in the growing spine”. The present paper groups participants’ varied experience according to principle and type of instrumentation, with a review of current studies of respiratory function following these types of surgery.

2. Posterior spinal distraction techniques

These techniques amount to an internal brace accompanying trunk growth while controlling spinal deformity, without, in principle, performing bone fusion. They are based on the first reports by Moe and Kharrat, using Harrington’s instrumentation [1].

2.1. Dual rod assembly

The principle consists in fitting 4 rods, subcutaneously or intramuscularly, with connectors on each side allowing iterative distraction at regular predetermined intervals (Fig. 2). The longest experience is that of the Growing Spine Study Group (GSSG), an international group of more than 30 experts, with results from a cohort of patients treated by growing rods since 1994. It is noteworthy that, since 2007, the group has ceased using single rods in the light of the advantages of dual rod assemblies. Current guidelines reserve this technique to children under 10 years of age, with curves exceeding 60°, after the family’s full informed consent to a long treatment program. Given the large number of operations required, non-operative treatment should be used to delay rod fitting for as long as possible, as each year reduces the overall complications rate by 13%. To avoid spontaneous fusion in the spine, rods should be positioned intramuscularly, under the superficial fascia, to limit skin impingement. Subperiosteal dissection during rod fitting should be avoided. Apical fusion, originally performed by some teams, is not recommended, unlike fusion of proximal and distal anchor points, which should use 4 fixation points on at least 2 adjacent vertebrae. Transverse connectors are unnecessary when fixation is provided by pedicular screws, but are preferable if hooks are used. The primary procedure and subsequent lengthening interventions should be performed under neurophysiological control and be followed by 6 months’ brace immobilization to achieve fusion at the extremities.

Mean intervals between distractions ranged between 10 and 20 months until 2003, but the tendency now is to keep them shorter, at

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**Fig. 1.** Current classification of early-onset scoliosis [9].

**Fig. 2.** Radiographs of different 2-rod posterior assemblies.
around 7 months: they should not exceed 6 months if gain in height is to approximate physiological growth (1.2–1.7 cm per year) [2]. Longitudinal follow-up of the cohort found a complications rate of 55%, with a mean of 2.2 complications per patient [10]. Fifty-eight percent of complications could be resolved during the following distraction, but unscheduled surgery was still required in 10% of patients. Dual rods considerably reduced the rate of mechanical implant failure, but at the cost of a slightly higher risk of infection [11]. Overall, the rate of complications per operation was around 18%, and each distraction procedure increased the complications risk by 24%. Actuarial survivorship modeling showed that a lengthening program initiated at the age of 6 years requires at least 10 lengthening interventions up to an age suitable for fusion, with a consequent complications risk exceeding 65%.

One of the most common complications is material breakage, with an incidence of 10% for bilateral rods. It usually occurs in the convex rod at the thoracolumbar junction, close to the connectors, which represent an area of increased rigidity [12]. No correlation was found between patient weight or type of implant (screw or hook). Thoracic hyperkyphosis, found in 30% of patients, on the other hand, is a major risk factor for mechanical failure, increasing overall complications risk during treatment by a factor of 3 [13]. Anchor-point failure is relatively rare following primary fusion, but incurs a risk of neurologic complications described by Skaggs et al., especially in case of intra-canal migration of proximal pedicular screws [14]. It is consequently recommended not to limit proximal fixation to a single pedicular screw on either side in long assemblies, but to add at least one hook or a second screw, to distribute anchorage stress.

The main problem with conventional growing rods is progressive spinal stiffness, with consequently diminishing growth in height as lengthening proceeds [15], the so-called “law of diminishing returns”: gain in Cobb angle is usually obtained only at the first procedure, with the fitting of the first distraction rods; thereafter, T1-S1 gain gradually decreases and more or less disappears after 6 distractions, or just 3 years of treatment at the optimal rhythm.

Bilateral growing rods are effective in early-onset scoliosis, stabilizing spinal deformity while achieving height gain close to that of physiological growth. Family compliance is essential in respecting distraction intervals and due to the high rate of complications. The major problem is progressive stiffening, found on average after 5 distractions, and partial or complete fusion, found in 80% of cases, hampering definitive fusion. Therefore, it is now recommended to continue non-operative treatment and delay rod fitting as long as possible.

2.2. Single-rod assembly

In France, the first unilateral assemblies were performed in the 1970s, using a subcutaneous Harrington rod. A brace was systematically associated. The intrinsic mechanical problems and need for iterative retensioning led to multiple proximal and distal fixations being introduced, with segmental instrumentation, and later to the introduction of a magnetic device (the Phenix rod) to avoid the need for iterative surgery. This device was abandoned in 2009 as unreliable, bulky and imposing a straight assembly on the spine. At the same time, the proximal and distal fixations were improved, leading finally to the instrumentation design used today [16,17].

The proximal fixation comprises 3 hooks (2 supralaminar and 1 pedicular) and distal fixation is provided by 2 pedicular screws, in what is known as H3S2 assembly [18,19]. The rod, in concave or in $ position, is curved according to the target spinal profile. Frontally, the vertical position increases biomechanical resistance to breakage. The rod is introduced intramuscularly. The soft tissue of the intermediate area must be respected to avoid any risk of stiffness or auto-fusion. The proximal and distal surgical approaches are limited.

Since 2005, 103 H3S2 assemblies have been performed. In some cases, depending on deformity severity, preoperative traction was used to improve flexibility, and the objective of the primary surgery was to maintain the resulting gain. Thirty-eight patients had at least 2 years’ follow-up. Mean Cobb angle was 65° preoperatively, 28° postoperatively and 32° at last follow-up; i.e., 54% improvement. Most patients showed improved profile. There were 7 proximal junctional kyphoses. Mean interval between retensioning procedures was 10 months. At last follow-up, 6 of the 38 patients (18%) had had at least 1 complication: 1 superficial infection, 2 deep infections that resolved after cleansing and antibiotherapy, and 4 rod breakages.

2.3. Magnetic growing rods

To avoid the drawback of surgical reintervention to lengthen the rod and follow physiological growth, rods were developed which could be lengthened by magnetic force applied transcutaneously. The only such system available today is MAGEC® (Ellipse Technologies, Inc.), comprising 4.5 or 5.5 mm diameter titanium motorized rods with 48 mm distraction reserve (Fig. 3). Contraindications include: age <2 years, weight <11 kg, BMI>25, pacemaker/defibrillator, and MRI required during the rod implantation period. Distraction is triggered by a programmable external magnet, with a dedicated device to locate the internal magnet. Lengthening is performed in consultation, without the need for anesthesia or analgesia, at a rhythm determined by the surgeon.

Benefit for patient, family and health system comprises [20]:

- fewer complications related to iterative surgery;
- less psychological trauma for patient and family;
- improved quality of life for child and family, with less time off school and off work;
- high primary outlay which is amortized within 3–4 years by savings on hospital stay.

The GES 2014 round-table reviewed files for 32 patients treated in France and Luxembourg using MAGEC® magnetic spinal growing rods for severe progressive scoliosis. The principal objective of this short follow-up preliminary study was to assess successful magnetic rod lengthening and Cobb angle stabilization. Thirty-two children, male and female, aged 5–11 years (mean: 9 years) were included in 6 centers. Forty-two rods were implanted (22 single, 10 dual). Mean follow-up was 13 months (range: 4–28 months). Rod lengthening was measured at each session and compared against the theoretic lengthening as set on the MAGEC® remote control.

A total of 118 lengthening procedures were performed. Mean programmed lengthening was 5.85 mm per session, and mean measured lengthening 3 mm. Twenty-one patients had satisfactory real lengthening (>80% of programmed value) at last follow-up. In 8 patients, real lengthening progressively decreased, due to progressive stiffening of scoliosis beyond the mechanical distraction capability of the rod. Three rods blocked and required replacement or abandonment of the technique. Mean Cobb angle was 72° preoperatively, 41° postoperatively and 45° at last follow-up.

Complications comprised: 5 proximal anchorage detachments in 3 patients, 2 rod breakages in 1 patient, 1 open scar exposing material, 1 deep infection requiring material ablation, and 1 superficial infection. Complications required 8 revision procedures.

This study confirmed the MAGEC® device’s technical lengthening capacity, only 7% of rods failing to function. The mean lengthening obtained, however, was almost 50% less than planned. This discrepancy may be explained by loss of force due to malpositioning of the magnetic field, an excessive thickness of soft tissue.
between field and rod, and above all insufficient magnetic field strength compared to the inevitable stiffening of the scoliosis over the period of lengthening, as was clearly found with conventional growing rods (so-called “law of diminishing returns”). The present study lacked sufficient follow-up to properly assess this stiffening, which would seem to be unavoidable.

Cobb angle showed clear improvement with primary surgery and then stabilized, but did not diminish over successive lengthenings. There is a mechanical limit to reduction imposed by the architectural deformity of the vertebrae and also by the solidity of anchorage, preventing the assembly progressively adapting to the changes in shape induced by lengthening.

The complications rate was comparable to that found with non-magnetic growing rods.

In conclusion, MAGEC® magnetic growing rods appeared to be mechanically reliable. However, their use associated with the rigid fixation currently available fails to prevent progressive spinal stiffening. The number of repeat procedures was significantly reduced, but the complications rate remained high.

2.4. VEPR

VEPTR (Vertical Expandable Prosthetic Titanium Rib) was initially developed for congenital thoracic and spinal deformities causing thoracic insufficiency syndrome, defined as the inability of the thorax to ensure normal respiration and good pulmonary development [21]. Such patients usually present “exotic” scoliosis [22], a rare, severe and complex subgroup of spinal deformity, often associated with deformed thorax due to the spinal deformity, vertebral rotation and dorsal lordosis or kyphosis, inhibiting thoracic and thus pulmonary growth [2,23]. Thoracic insufficiency may be aggravated by associated costal abnormalities, leading to thoracic cage rigidity.

VEPTR is a longitudinal costal expansion implant, comprising 1 costo-costal and 1 hybrid component (rib/lumbar hook or rib/pelvis). The principle, rather than correcting spinal deformity, is to enlarge the narrow thorax so as to create an expansion chamber allowing the lungs to develop and thereby correct the thoracic insufficiency syndrome. The main indications are: thoracic deformity impairing thoracic function, such as congenital scoliosis with unilateral segmentation disorder and rib fusion (e.g., spondylocostal dysplasia [24]); unstable thorax due to a missing rib; bilateral rib fusion (e.g., Jarcho-Levin syndrome [25]); short narrow thorax (e.g., Jeune syndrome [26]); or, by extension, severe progressive infantile scoliosis. The cost is expensive compared to a classical assembly using implants derived from the Cotrel-Dubousset instrumentation.

Like the other fusionless spinal distraction techniques, VEPTR theoretically requires expansion procedures every 4–6 months to enlarge and increase the height of the hypoplastic thorax, stabilize the wall defect and try to progressively improve associated spinal deformities. Opening thoracotomy may be needed in rib fusion areas or for missing ribs, with resection of pleural fibrosis. Experience shows that costal synostoses recur, requiring iterative costotomy at each distraction procedure (Fig. 4).

VEPTR is used in a wide variety of etiologies apart from its primary indication for thoracic insufficiency, making it difficult to assess the published results and the impact on respiratory function.

As with fusionless distraction techniques using single or dual rods, initial angular gain subsequently stagnates or diminishes [27]. Moreover, the complications inherent to fusionless spinal distraction surgery are frequent. A recently published review of the French VE PTR series reported a high complications rate, comparable to other series [28], although, with experience, recommendations can be made to limit the incidence.

Thus, more precise assessment of this technique would have to be based on the indications for which it was initially designed: thoracic insufficiency syndrome and malformative scoliosis with associated costal abnormality. Assessment, if it is to determine efficacy, should include not only vertebral and costal deformity at follow-up but also respiratory function, which is difficult to explore in small children. Even so, VEPTR is a means of optimizing the
management of patients with major trunk deformity, sometimes at the limit of therapeutic possibility.

3. Growth guidance

Two growth guidance techniques are currently most widely used: the Shilla technique [5] and Modern Luque Trolley rods [4].

3.1. Shilla

The principle of the Shilla instrumentation, described by McCarthy in 2008, is to control the scoliosis by guiding growth with a single posterior procedure, without iterative surgery or brace [5]. The assembly uses 2 rods fixed to 3 or 4 pedicular screws inserted in the apical vertebrae, with fusion of the summit of the curve; pedicular screws are inserted extra-periosteally in the proximal and distal extremities of the rods, enabling the rods to slide, guiding spinal growth (Fig. 5).

For this round-table, 11 prepubertal children aged 4 years 6 months to 11 years 6 months were reviewed at a mean 3 years’ follow-up (range: 2–4.5 years). Six had undergone definitive fusion by end of follow-up. The number of instrumented vertebrae ranged from 11 to 14. Mean initial Cobb angle was 57°. Five had 1 and 6 had 2 unscheduled procedures. Four curves showed good control (57° Cobb angle corrected to 42°) and 7 failed to maintain correction (57° angle unchanged). In the 4 children free of complications, seated height increased from 67 cm to 72.5 cm; in McCarthy’s series, it increased by a mean of 12%. Global angle was less well controlled than in McCarthy’s series (70° corrected to 34°). Seven of the 11 children had unexpected complications, compared to 50 in McCarthy’s series. The high rate of material disassembly was due to poor screw fixation in small pedicles, with excessive stress; this explains the metallosis found on revision surgery. A crankshaft phenomenon was found in 7 children, both at the curves and at the fused summit; these problems occurred in children with a mean age of 10 years 9 months: i.e., at pubertal growth, which overpowered the instrumentation, despite fusion of the summit. The 4 children with well-controlled scoliosis had a mean age of 7 years 8 months, before the dangerous pubertal growth peak; many children in McCarthy’s series were still far from this age. Surgery was aggressive, due to the size of the approach, the length of surgery time (4h 54 min) and quantity of blood loss (425 cc for McCarthy).
4. Growing rods in neuromuscular pathology

Fusionless instrumentation in neuromuscular scoliosis concerns patients who are often malnourished, with poor quality bone and exposure to frequent complications such as implant detachment or protrusion. Preoperative nutritional, respiratory or orthopedic preparation (by progressive traction via a cranial halo) facilitates surgery and limits risk.

To reduce the rate of complications, and of mechanical complications in particular, a certain number of guidelines are to be respected. Spinal fixation should use multiple solid anchors. Fixation should not be onto the ribs, which are fragile, but rather onto the vertebrae, with supra- or sub-laminar hooks, which resist detachment better than screws. Proximal anchorage should extend up to the first thoracic or even last cervical vertebrae, to avoid the risk of proximal junctional kyphosis. In non-walking patients, distal fixation should be supported by the pelvis, with solid, stable, small anchorage. Pelvic fixation by iliosacral screws meets these requirements [29,30]. Spinal assembly should be fairly symmetrical, to neutralize balance disorder in the trunk and pelvis and distribute stress. This means it must be bilateral, supported by the pelvis (Fig. 6). To reduce operative risk in fusion, surgery should be as non-invasive as possible, restricting the approach to two short incisions over the anchor points.

Fusionless instrumentation is indicated in neuromuscular scoliosis in case of failure, impossibility or difficulty of non-operative treatment or of contraindications to fusion due to fragile diathesis. The indication should be early, before onset of puberty, while the spinal deformity is still supple and reducible. The benefits of fusionless instrumentation in neuromuscular scoliosis are, in our experience, multiple: improved health and functional status and improved quality of life for both patient and family.

5. Definitive fusion surgery

A key point in treatment is the transition from distractions to definitive fusion. Until recently, the literature was sparse, but Flynn et al. reported a series of 99 patients, aged 11–13 years, undergoing lengthening over a mean of 5 years [31].

Definitive fusion was indicated either because the patient had reached maturity, or because spinal stiffening was so severe that distraction no longer provided benefit, or again because of mechanical or infectious complications. The fusion levels were the same as for lengthening rods in 55% of cases, but extension to a further 1 or 2 levels, usually distally, was required in respectively 15% and 9% of cases. Proximal extension was usually due to mechanical failure or proximal junctional kyphosis, while distal extension was more often to improve frontal balance. Intra-operative findings included difficulty of exposure, a mean 3-fold more bleeding than in classical fusion, and spontaneous fusion in 62% of cases. The fused anchorages were generally reused, changing implants. The rate of neurologic complications was 3%, with favorable evolution in all cases. Due to stiffening or complete fusion, the number of implants was usually small, and frontal correction was limited: <50% in almost 75% of cases, despite frequent (24%) use of osteotomy. The authors also highlighted the difficulty of sagittal correction in these spines with little mobility.

6. Respiratory function results

6.1. Reminders

During postnatal lung growth, alveolae multiply from 20–50 million in neonates to 300 million by the age of 2 years. Their size then increases, doubling between 2 years of age and adolescence.
Thoracic volume increases, reaching 30% of adult volume by 6 years and 50% by 10 years of age [33]. Compliance is high in the infant thorax, and decreases by 30% between the ages of 5 and 16 years.

Respiratory function study requires cooperation, and is not feasible until 5 or 6 years of age. Assessment criteria essentially comprise forced vital capacity (FVC) and maximal expiratory volume per second (MEVS). Thoracic insufficiency syndrome, a concept introduced by Campbell, is defined as the inability of the thorax to ensure normal respiratory function and pulmonary growth [34]. It may be of thoracic or spinal origin. It is not determined by any objective quantitative value.

6.2. Influence of scoliosis

Depending on severity, scoliosis may induce respiratory restriction syndrome. For Cobb angles exceeding 100°, FVC is reduced by 30%; above 120°, there is a risk of respiratory failure and chronic pulmonary heart disease. Scoliosis induces a specific respiratory profile, with reduced current volume and superficial polynema, increased respiratory work and a 3-to-5-fold increase in the energy cost of tissue oxygenation. In neuromuscular scoliosis, there is poor correlation between Cobb angle and the severity of the restriction syndrome, due to multiple associated factors.

6.3. Effect of distraction

Olson et al. studied the effect of costal distraction in an experimental animal model of costal synostosis in rabbit [6]. Synostosis was induced at 5 weeks of life, followed in one group by resection and implantation of a thoracic distractor at 10 weeks; healthy animals constituted the third (control) group. Pathologic examination of the pulmonary parenchyma at 24 weeks of life found, in the synostosis-only group, larger alveolar volume, emphysema and reduced vessel size; in the synostosis + distraction group, vascularization was considerably better, suggesting a positive remodeling effect. Motoyama et al. studied the effects of VEPTR distraction at 3 years' follow-up in 24 children with congenital scoliosis, aged 2–11 years [7]. FVC increased by a mean of 11% per year, particularly in the younger patients (< 6 years). On the other hand, there was a 44% decrease in thoracic compliance, due to parietal stiffening. Mayer et al., in a multicenter prospective study of 53 patients aged 4–15 years treated by VEPTR for thoracic insufficiency syndrome, found reduced FVC and MEVS despite significant improvement in Cobb angle (from 95° to 61°) by 8 months' follow-up. The same team more recently showed an absence of correlation between Cobb angle and FVC in 10 children with infantile or congenital scoliosis under 3 years of age [8]. Moreover, in 15 patients aged 2–10 years managed by VEPTR, they found no correlation between improvement in Cobb angle and evolution of FVC at 13 months' follow-up. Yoon et al., in 6 children with neuromuscular scoliosis aged 5–10 years, reported the effect of spinal distraction by MAGEC® magnetic rods on respiratory function [35]; at 2 years' follow-up, Cobb angle showed 34% improvement, FVC 14% and MEVS 17% over initial values. These results may have been due to early surgery and iterative non-invasive lengthening, limiting fibrous parietal retraction. However, the series was small and the study involved several biases; the results need confirming.

The increase in thoracic volume following costal distraction is not accompanied by improved respiratory function. Relations are complex and associated factors numerous. Motorized vertebral and costal distraction reduces fibrosis and thus thoracic stiffness, and may in future offer a solution.

7. Conclusion

Surgical techniques that conserve spinal growth are effective in controlling early scoliosis and to some extent restoring spinal growth, but show a high rate of complications. They require multi-disciplinary teams with specific experience, given all the associated abnormalities such as respiratory disorder and denutrition. Gain in respiratory function is harder to assess: increased thoracic volume allowing pulmonary development does not systematically lead to improved respiratory function, particularly due to decreased thoracic compliance. For all these reasons, these techniques should
be used only after exhausting all the possibilities of non-operative management.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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