



Actual evidence in the medical approach to adolescents with idiopathic scoliosis

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Idiopathic scoliosis (IS) is a three-dimensional deformity of the spine and trunk. The most common form involve adolescents. The prevalence is 2-3% of the population, with 1 out of 6 patients requiring treatment of which 25% progress to surgery. Physical and rehabilitation medicine (PRM) plays a primary role in the so-called conservative treatment of adolescents with IS, since all the therapeutic tools used (exercises and braces) fall into the PRM domain. According to a Cochrane systematic review there is evidence in favor of bracing, even if it is of low quality. Recently, a controlled prospective trial including a randomised arm gave more strength to this conclusion. Another Cochrane review shows that there is evidence in favor of exercises as an adjunctive treatment, but of low quality. Three meta-analysis have been published on bracing: one shows that bracing does not reduce surgery rates, but studies with bracing plus exercises were not included and had the highest effectiveness; another shows that full time is better than part-time bracing; the last focuses on observational studies following the Scoliosis Research Society (SRS) criteria and shows that not all full time rigid bracing are the same: some have the highest effectiveness, others have less than elastic and nighttime bracing. Two very important RCTs failed in recruitment, showing that in the field of bracing for scoliosis RCTs are not accepted by the patients. Consensuses by the international Society on Scoliosis Orthopedic and Rehabilitation Treatment (SOSORT) show that there is no agreement among experts either on the best braces or on their biomechanical action, and that compliance is a matter of clinical more than patients' behavior (there is strong agreement on the management criteria to achieve best results with bracing). A systematic review

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of all the existing studies shows effectiveness of exercises, and that auto-correction is their main goal. A systematic review shows that there are no studies on manual treatment. The SOSORT Guidelines offer the actual standard of conservative care.

KEY WORDS: Adolescents - Idiopathic scoliosis - Rehabilitation medicine.

Idiopathic scoliosis (IS) is a three-dimensional deformity of the spine and trunk.¹ The most common form involve adolescents (*adolescents idiopathic scoliosis*, AIS). The prevalence for AIS is 2-3% of the population, with 1 out of 6 patients requiring treatment of which 25% progress to surgery. Physical and rehabilitation medicine (PRM) plays a primary role in the so-called conservative

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treatment of adolescents with IS (AIS), since all the therapeutic tools used (exercises and braces) fall into the PRM domain. The knowledge of the actual evidence is the only possible base for a correct approach to these patients.

According to the actual definition, evidence based clinical practice means combining the actual best evidence with clinical expertise and patients' preferences.¹ Theoretically, treatments should be applied only when there is evidence of their effectiveness, but this is not the reality with traditional treatments established for many years. In fact, Clinical Guidelines give their recommendations according to level of evidence, which must be well defined.² The so-called pyramid of evidences is described: the highest level of evidence is achieved by meta-analysis of RCTs and Cochrane reviews, and the lowest by Clinical Experts' Consensuses. Nevertheless, the lowest can sometimes be the actual best or the only achievable evidence. Consequently, it is accepted that, while waiting for higher levels of evidence, clinical decisions should be driven by the actual scientific knowledge, even if it is not (yet) of the highest standard: in these cases a low quality of evidence is declared.

Another point well accepted by modern evidence based medicine is that not in all fields it is possible to perform randomized controlled trials (RCTs). In BMJ, a meta-analysis (*i.e.*, a study with the highest level of evidence) showed that it is not possible to find any evidence on the effectiveness of parachutes while falling from airplanes, since there are no RCTs.³ This was a nice paradox to explain situations in which RCTs are not appropriate.

Finally, it is known that if RCTs are not possible for any reason, observational studies are a way to achieve good quality evidence.^{4,5} Moreover, RCTs represent experimental settings (*i.e.*, they lack good ecological reliability, since they do not fully represent the everyday clinical life due to their usual strict inclusion criteria): observational studies overcome this limitation and should always be performed after RCTs to verify their real applicability.

According to all these premises, the aim of this paper was to review the actual best evidence on the PRM approach to adolescents with IS, reviewing the best papers published on the topic that formed the base of the Clinical Guidelines pub-

lished by the international Society On Scoliosis Orthopedic and Rehabilitation Treatment (SOSORT).¹

Evidence on bracing

According to a Cochrane systematic review there is evidence in favor of bracing, even if it is of low quality.⁶ Due to the paucity of RCTs in the literature, this review included RCTs and quasi-RCTs (QRCT), *i.e.*, prospective controlled cohort studies. One QRCT⁷ with 286 girls curbed curve progression at the end of growth (success rate 74% [95% CI: 52% to 84%]), better than observation (success rate 34% [95% CI: 16% to 49%]) and electrical stimulation (success rate 33%, 95% CI: 12% to 60%): since it was a QRCT, the quality of evidence was judged very low. Moreover, one RCT⁸ had low quality evidence. It included 43 girls and concluded that a rigid brace is more successful than an elastic one (SpineCor) at curbing curve progression when measured in Cobb degrees: this study did not find significant differences between the two groups in the subjective perception of daily difficulties associated with wearing the brace. After the publication of this Cochrane Review, another very important study was published in 2013,^{9,10} a QRCT with a randomized arm: the National Institute of Health (NIH) Ethical Committee required the trial to be stopped early, owing to the efficacy of bracing. Treatment was successful in 72% of cases after bracing, *versus* 48% after observation (propensity-score adjusted odds ratio for treatment success 1.93, 95% CI 1.08-3.46). Also an intention-to-treat analysis was performed in the randomized arm: treatment succeeded in 75% braced patients *versus* 42% observed (odds ratio 4.11, 95% CI 1.85-9.16). Moreover, it was found a positive association between hours of brace wear and rate of treatment success ($P < 0.001$). Consequently, the actual evidence is in favor of bracing, but it is of low quality, since it is not based on a RCTs. Similarly, there is evidence of superiority of rigid *versus* elastic bracing, but of low quality. Until another, higher level of evidence is offered, clinicians should act according to this best evidence.

About future possible evidence, it must be stated that already two very important RCTs failed in recruitment, showing that in this field RCTs are not well accepted by the patients.¹¹⁻¹³ The first paper

comes from an RCT attempted in Holland.¹¹ A total of four patients were included, and 14 refused to participate in a 18-month period. There were a lot less eligible patients than anticipated (40 instead of 100 per year), and the patients' participation rate was much lower than in a previous pilot study (21% instead of 70%).¹¹ Consequently, the RCT failed because of an overestimation of the number of eligible patients and because a lot less of eligible patients were willing to participate compared to the previous pilot study. The author concluded that "a reason for the low participation rate could be that this trial evaluated a frequently used existing treatment instead of a new treatment, and patients and parents might be afraid of not being treated (despite an intensive secure system for the control arm)".¹ The second paper is the QRCT already cited.^{9, 10} This paper started as an RCT, but failed as such.^{9, 10, 12} Out of 1086 eligible patients (100%), 703 declined to participate (64.7%), and 155 accepted to be randomized (14.3%): in the final study, 116 patients (10.7%) were included and underwent randomization, but 10 did not follow it. In the end, out of 1086 patients, only 9.8% (106) were correctly randomized and included: this result points out the difficulties of RCTs in this field. In fact, patients and parents perceive the bracing RCTs as parachute trials³ and prefer a shared decision with their physicians. Classical RCTs cannot be performed. Observational trials are a viable alternative to RCTs, but they should mainly focus on the SRS methodological criteria for bracing research.⁶

A meta-analysis showed that bracing does not reduce surgery rates,¹³ but studies with bracing and exercises were not included and had the highest effectiveness.¹⁴⁻¹⁶ In fact, it was performed on English language clinical papers including observation or a TLSO (without any adjunctive treatment) in sample closely matching the current indications for bracing (skeletal immaturity, age <15 years, Cobb angle between 20° and 45°). Eighteen Level III or IV clinical series (observation=3, bracing=15) were included. There was some uniformity in surgical indications, but the surgical rates were extremely variable: from 1% (out of 72 patients) to 43% (out of 120) after bracing, from 13% (out of 15) to 28% (out of 47) after observation. When pooled, the bracing surgical rate was 23% compared with 22% in the observation group. Four papers by SOSORT

members⁶⁻⁸ (all excluded because exercises had been added to bracing), reported in the same population surgery rates between 2% and 7% according to an efficacy analysis (similar to that of the meta-analysis). In two of these papers (Level II prospective studies) also an intent-to-treat analysis has been performed, with surgery rates of 12% and 14%.^{7, 8} These data question the generalizability of this review out of a US/Northern Europe settings, and strengthen the idea of effectiveness of exercises as an adjunctive, but also as an adjuvant treatment to bracing.

Another meta-analysis shows that full time is better than part-time bracing.¹⁷ The members of the Prevalence and Natural History Committee of the Scoliosis Research Society (SRS) conducted a meta-analysis of 20 studies (1910 patients): 1459 braced, 322 treated with lateral electrical surface stimulation (LESS), and 129 observed. The weighted mean proportion of success were: 0.39 for LESS and 0.49 for observation; for bracing it ranged from 0.60 (8 hours per day - h/d), to 0.62 (16 h/d) to 0.93 (23 h/d). The 23 h/d were significantly more successful than any other treatment ($P < 0.0001$) or bracing for 8 or 16 h/d ($P < 0.0001$), while the difference between the 8 and 16 h/d was not significant. The weighted mean proportion of success for the six types of braces included in this review was 0.92, with the highest proportion (0.99) achieved with the Milwaukee brace. Even if this paper is quite old, it gives some useful insights still valid.

A meta-analysis (now under review) of observational Level II and III studies following the SRS methodological criteria for research on bracing,¹⁸ shows that not all full time rigid bracing regimen are the same: some have the highest effectiveness, others have less than elastic and night-time bracing. It found 9 papers (2 excluded for very low quality), 1698 patients (551 included). After 40.5 months of treatment (range 16.4-70.8) the results were: 23.4% (range 0-78) progressed more than 6° Cobb, 13.4% (0-54) finished treatment above 45°, and 21.6% (0-71) were fused; 62.5% of the patients reached a 2 years follow-up, and the total fused was 24%. Striking differences have been found in subgrouping with either the best results or the worst results in full-time rigid bracing: the best results were in studies following the SOSORT criteria, with exercises added, and with treatment lasting more than 50 months. Intermediate among

these two groups were the results of elastic and night-time bracing. This paper gives indications about the actual predictive factors of good results (*i.e.*, best clinical behaviors) in the worst clinical situation (patients 25-40° Cobb, Risser 0-2).

SOSORT Consensus shows that there is no agreement among experts, either on the best braces, or on their biomechanical action.^{19, 20} This paper was produced through a Delphi Procedure, with final Consensus Conference among SOSORT members. The Chêneau brace was the most frequently recommended. The importance of the three-point system mechanism was stressed. Options about proper pad placement on the thoracic convexity were divided: 50% for the pad reaching or involving the apical vertebra and 50% for the pad acting caudal to the apical vertebra. There was agreement about the direction of the vector force, 85% selecting a “dorso lateral to ventro medial” direction but not about the shape of the pad to produce such a force. Principles related to three-dimensional correction achieved high consensus (80-85%), but suggested methods of correction were quite diverse. This first Consensus study reveals that there continues to be a strongly held and conflicting, if not a contentious, opinion regarding brace design and treatment. Consequently, it is not possible today to define the best brace, and the best biomechanical corrective approach.

Another SOSORT Consensus states that compliance is a matter of clinical more than patients' behavior and there is strong agreement on the management criteria to achieve best results with bracing.^{1, 19} Also this paper followed a Delphi Procedure with final Consensus Conference among SOSORT members. 90% agreement was set as the minimum to be reached. A final set of 14 recommendations was given, grouped in 6 domains (experience/competence, behaviours, prescription, construction, brace check, follow-up). With increasing experience in bracing by SOSORT members, all numerical criteria tended to become more strict. SOSORT recommends to professionals engaged in patient care to follow the Guidelines of this Consensus in their clinical practice. The SOSORT criteria should also be followed in clinical research studies to achieve a minimum quality of care. It is possible to define the best management strategies to help patients achieve a good compliance and perform the best treatment. These strategies can be

resumed in specific medical and technical expertise, team approach and clinical behaviors.

Physiotherapeutic specific scoliosis exercises (PSSE)

A Cochrane review shows that there is evidence in favor of exercises as an adjunctive treatment, but of low quality.²¹ It focused on RCTs and QRCTs, including two studies (154 participants). One RCT²² showed that exercises as an adjunctive to other conservative treatments increase the efficacy of these treatments (thoracic curve reduced: mean difference 9.00, 95% CI 5.47-12.53; lumbar curve reduced: mean difference 8.00, 95% CI 5.08-10.92): due to the methodological characteristics of the study, it has been considered low-quality evidence. Moreover a QRCT²³ (very low-quality evidence) almost proved that PSSEs can reduce brace prescription (risk ratio 0.24, 95% CI 0.06-1.04) as compared with “usual physiotherapy” (general exercises according to the preferences of the single therapists within different facilities): in fact, in the original study the statistical significance was reached, while the different statistical analysis performed by the Cochrane authors did not reach significance for a lack of power. This Cochrane review shows that there is evidence in favor of exercises effectiveness, even if of low quality.

Repeated systematic reviews of all the existing studies showed the effectiveness of PSSE and that auto-correction is the main goal.²⁴⁻³⁰ Nineteen studies were retrieved,²⁷ including one RCT and eight controlled studies; 12 studies were prospective. There were 1654 treated patients and 688 controls. Three papers on Scoliosis Intensive Rehabilitation (Schroth), five on extrinsic autocorrection-based methods (Schroth, side-shift), four on intrinsic autocorrection-based approaches (Lyon and SEAS) and five with no auto-correction (three asymmetric, two symmetric exercises) were found. Apart from the oldest and technically less reliable paper (no auto-correction, symmetric exercises, very low methodological quality), all studies confirmed the effectiveness of exercises in reducing the progression rate (mainly in early puberty) and/or improving the Cobb angles (around the end of growth). Exercises were also shown to be effective in reducing brace prescription. PSSEs are

based on the auto-correction and have the highest effectiveness.

Other conservative treatments

A systematic review has shown that there are no studies on manual treatment.³¹ It focused on any kind of research on AIS patients treated exclusively by chiropractic manipulation, osteopathic techniques, massage, with outcome in Cobb degrees. Only three papers were found. However, no one of the three satisfied all the required inclusion criteria because they were characterized by a combination of manual techniques and other therapeutic approaches. This paper, and the meta-analysis by Rowe¹⁷ showing no effectiveness of electrical stimulation, excludes evidence on other conservative treatments to control the curve evolution.

Conclusions

Research on conservative treatment of AIS has continuously decreased since the 1980s.^{32, 33} SOSORT is born as a cause (or effect) of the renewed interest on research in this field begun with the new millennium. The SOSORT Guidelines offers the actual standard of conservative care.¹ They are totally evidence-based and shows that the existing evidence on conservative treatment today is low: out of 65 recommendations, no one was Level I (strong evidence), 2 were Level II, the remaining were lower. Nevertheless, when importance for patients is considered, 13 were Grade A (to be applied to all patients), and 49 grade B (almost all patients). The correct answer to this situation is research and increased efforts to search for evidence.

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