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To screen or not to screen for adolescent idiopathic scoliosis? A review of the literature



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ABSTRACT

Objectives: Over the years, there has been much debate about the desirability and efficacy of screening for adolescent idiopathic scoliosis. To analyse the current evidence from a public health point of view a renewed evaluation of the literature was performed.

Study design: Literature review.

Methods: We performed two literature searches: from January 2000 to April 2015 for systematic reviews or guidelines on screening for adolescent idiopathic scoliosis and from January 2009 to April 2015 for all studies on adolescent idiopathic scoliosis and screening methods. We evaluated if screening for adolescent idiopathic scoliosis fulfils the criteria of the UK National Screenings Committee for appraising a screening programme.

Results: Adolescent idiopathic scoliosis is a condition with an unpredictable natural history. The optimal age and frequency of screening are unknown; it is not possible to predict which patients will need (conservative or surgical) treatment. The Forward Bending Test has a positive predictive value of 2.6% and a sensitivity of 56% (95% CI 23–88%) for (conservative or surgical) treatment, and is therefore not valid enough for use in a screening programme. There seems to be sufficient evidence that brace treatment is effective for preventing progression of adolescent idiopathic scoliosis. It is not clear if screening is cost effective.

Conclusions: Despite evidence that brace treatment is effective for preventing progression of adolescent idiopathic scoliosis, the benefits from the screening programme do not outweigh the harms. From a Public Health point of view, there is not enough evidence to support a screening programme for adolescent idiopathic scoliosis.

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Introduction

Adolescent Idiopathic Scoliosis (AIS) is a deformity characterised by a three-dimensional curvature of the spine; if all small curvatures are included it affects 2–3% of adolescents. The curvature is determined by measuring the Cobb angle on

an X-ray of the spine. Internationally, a scoliosis is diagnosed if the Cobb angle is $\geq\!10^{\circ}.^{1}$

Over the years, there has been much debate about the necessity and efficacy of screening adolescents for AIS.^{2–6} The most important points of criticism were the accuracy of the screening test, the unpredictable natural history and insufficient evidence for brace treatment. One of the most recent

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reviews presented statements and recommendations based on a combination of a literature review (search period until 2010) and expert opinion.⁷

Recently, the results of a randomized controlled trial (RCT) were published, with new evidence for the effectiveness of brace treatment for AIS.⁸ To analyse the current evidence from a public health point of view, we considered a new evaluation of the literature to be desirable.

The UK National Screening Committee (UKNSC) has formulated criteria for appraising the viability, effectiveness and appropriateness of a screening programme. Ideally, all of these criteria should be met before screening for a condition is initiated. These criteria are based on Wilson and Jungner's classic criteria to guide the selection of conditions that would be suitable for screening.

In this article, we will evaluate if AIS fulfils the criteria of the $\ensuremath{\mathsf{UKNSC}}$.

Methods

The following databases were searched from January 2000 until April 2015 for systematic reviews or guidelines on screening for AIS: Guidelines International Network, National Institute for Health and Care Excellence, Cochrane Reviews, United States Preventive Services Taskforce.

We also performed a search in PubMed from January 2009 until April 2015. The following search terms were used: adolescent AND scoliosis AND idiopathic, combined with (using 'AND' and 'OR') 'predictive value of tests', 'mass screening', 'scoliosis/diagnosis', 'screening', 'screening tests', 'early diagnosis', 'Adam's forward bending test', 'forward bending test'. A total of 427 articles were found, of which 23 were relevant to the current review. Using the 'snowball method', we used the reference list of articles to search for other relevant articles.

We selected articles about screening methods that are suitable for use in public health settings, i.e. methods that are non-invasive, require little time or special equipment. The consensus statement of Labelle et al. concluded that the scoliometer used on a Forward Bending Test (FBT) is currently the best available technique for scoliosis screening. Moiré topography is another available screening technique, but requires special equipment, interpreting the results can be challenging and it takes more time. Therefore, we narrowed our search to literature on the FBT and scoliometer.

The criteria formulated by the UKNSC are divided into four categories: the condition, the test, the treatment, and the screening programme. The literature found was used to determine if the criteria of the UKNSC are met. Criteria regarding genetic screening are not discussed in this article because they are not relevant to the subject.

Results

The condition

1. The condition should be an important health problem.

In the general population, the prevalence of scoliosis with a Cobb angle $\geq 10^{\circ}$ is approximately 2–3%. However, the reported incidence of severe curves (Cobb angle $\geq 30^{\circ}$) varies from 0.01 to 0.3%. $^{10-13}$ If a curve shows progression and the Cobb angle is between 25° and 45° (with skeletal immaturity), brace treatment is indicated. If further progression occurs despite brace treatment and if the Cobb angle is $\geq 45^{\circ}-50^{\circ}$ (with skeletal immaturity), surgical treatment is indicated. 8,14 If, after complete skeletal maturation, the Cobb angle is $\geq 30^{\circ}$, there is some increased risk for problems in adult life, such as reduced quality of life, pain, functional impairments and sometimes pulmonary problems. 15,16 Depending on the inclusion criteria used, approximately 10% of patients require brace treatment and approximately 0.1–0.3% require surgical treatment. 1

The epidemiology and natural history of the condition, including development from latent to declared disease, should be adequately understood and there should be a detectable risk factor, disease marker, latent period or early symptomatic stage.

Five percent of the patients with a Cobb angle of $>10^{\circ}$ shows progression to $>30^{\circ}$.^{17,18} In earlier studies the possibility of improvement of the curve has been described.^{19,20} This has not been found in the more recent literature.¹

The cause of AIS is unknown; it probably has a multifactorial aetiology. It is not possible to make a reliable prediction as to which curves will show progression.

3. All the cost-effective primary prevention interventions should have been implemented as far as practicable.

Currently, there are no evidence-based primary prevention programmes.

The test

4. There should be a simple, safe, precise and validated screening test.

There are several tests that can be used for screening for AIS. The Adam's forward bending test (FBT) and scoliometer are suitable for screening purposes, because of the relatively little time and equipment that is needed. These tests are not diagnostic, X-rays are necessary to diagnose (the severity of) scoliosis. Currently, the scoliometer is the best tool available for scoliosis screening.⁷

In a meta-analysis of 36 studies investigating the clinical effectiveness of screening for AIS positive predictive values (PPV) of 28%, 5.6% and 2.6% were found for curves of >10°, >20° and (brace or surgical) treatment, respectively. None of the studies reported the specificity of screening for AIS. Only one study reported the sensitivity of the screening programme: Yawn et al. investigated the effectiveness of screening for AIS in grade 5–9 by means of the FBT combined with the scoliometer. They found a sensitivity of 64% (95% CI 45%–83%) for curves \geq 20° and 56% (95% CI 23%–88%) for treatment. The PPV of 2.6% for treatment means that, of 1000 adolescent referred for suspected scoliosis, 26 will be treated with

brace or surgery. A significant part of the remaining adolescents will have to be monitored. The sensitivity of 56% for treatment means that, of the 100 adolescents needing treatment (sometime in the future), 44 will be missed by screening.

Fong et al.²¹ also showed that studies using only the FBT had a higher referral rate compared to studies using FBT in combination with other screening methods (like the FBT combined with scoliometer or Moiré topography): 7.2% against 2.6%. Compared to studies using other screening methods, screening with only the FBT showed a lower PPV for curves >10° (23.2% vs 38%) and >20° (3.5% vs 11.0%).²¹

5. The distribution of test values in the target population should be known and a suitable cut-off level defined and agreed.

For the FBT, there are no referral values that have both a sensitivity and a specificity that are high enough for use in a screening programme. Several studies have tried to determine the optimal cut-off point for referral when using the FBT and the scoliometer. However, choosing a high specificity will negatively influence the sensitivity and vice versa. Therefore, Pruijs et al. suggested determining a range within which the examination should be repeated after 3–6 months. ²³

6. The test should be acceptable to the population.

As far as we know, there is no research on this subject. The screening programmes that were performed in the 1980's—1990's were acceptable in those days. Because of the non-invasive nature of the test we expect that the FBT is acceptable to the population.

There should be an agreed policy on the further diagnostic investigation of individuals with a positive test result and on the choices available to those individuals.

If the screening test is positive, an X-ray of the spine will be made to determine the Cobb angle. This is currently the gold standard for diagnosing scoliosis. For referral values, see criterion 5.

The treatment

 There should be an effective treatment or intervention for patients identified through early detection, with evidence of early treatment leading to better outcomes than late treatment.

The main conservative treatment options for the prevention of scoliosis progression include scoliosis-specific exercises (SSE), other forms of general physical therapy, and bracing.²⁴

The use of exercises or general physical therapy for the treatment of AIS is controversial.²⁵ A Cochrane review concludes that there is a lack of high quality evidence to recommend the use of SSE for AIS.²⁵

The objective of brace treatment is to prevent further progression of the curvature by means of external forces and, in some designs, the stimulation of active correction as the patient moves the spine away from pressures within the brace. The effect of bracing has been the subject of discussion for years. A Cochrane review on the effect of brace treatment found very low quality evidence in favour of using braces, making generalization very difficult.²⁶

Recently, the results of an RCT to determine the effectiveness of bracing compared with observation were published.8 The study was designed as a randomized trial, but fewer families than expected accepted randomization because of a preference for brace treatment. Therefore, a preference group was added to the trial: patients that were eligible for randomization but chose brace treatment. Inclusion criteria were: an age of 10-15 years, no previous treatment for AIS, skeletal immaturity (defined as a Risser grade [a measure of the amount of ossification and eventual fusion of the iliac apophysis, on a scale of 0-5, with higher grades indicating greater skeletal maturity] of 0, 1, or 2), and a Cobb angle for the largest curve of 20°-40°. Treatment success was defined as skeletal maturity without curve progression to \geq 50°, treatment failure was curve progression to \geq 50°. The study was stopped early because of the positive results. In the intention-to-treat-analysis of the randomized patients, the rate of treatment success was 75% after bracing and 42% after observation (OR 4.1; 95% CI 1.85-9.16). The number needed to treat (NNT) to prevent one case of surgery was 3.0 (95% CI 2.0-6.2). Longer hours of brace wear were associated with greater benefit. The authors conclude that bracing significantly decreases the progression of high-risk curves to the threshold for surgery in patients with AIS.

An important aspect for the effectiveness of brace treatment is brace wear compliance. There appears to be a positive correlation between compliance and the effect of the treatment. Second 27.28 Compliance is often not optimal; braces are worn 45–75% of the prescribed time. In an older publication, braces were worn only 10% of the prescribed time. It is probable that in general clinical practice the compliance is even lower than in the setting of a study, the awareness of being monitored seems to increase wearing time. This means that the effectiveness of brace treatment in general clinical practice might be lower than described in the study of Weinstein et al.

There should be agreed evidence based policies covering which individuals should be offered treatment and the appropriate treatment to be offered.

See above, criterion 8.

10. Clinical management of the condition and patient outcomes should be optimised in all healthcare providers prior to participation in a screening programme.

Based on the current search, we cannot answer the question if this criterion is met.

The screening programme

11. There should be evidence from high quality Randomised Controlled Trials that the screening programme is effective in reducing mortality or morbidity.

There are no RCT's on the effectiveness of screening for AIS. Bunge et al. performed a study that comes close to the level of evidence of an RCT.5 With a case-control study the effectiveness of screening for AIS was investigated by comparing 108 surgically treated patients to 216 healthy controls without AIS, matched for age and gender. With this study the effectiveness of the whole course of screening was investigated, from screening test to diagnostics and treatment.5 The authors investigated if cases and controls were screened for AIS. Patients detected by screening were younger at the time of diagnosis compared to otherwise detected patients (mean 10.8, SD \pm 2.6 vs 13.4 SD \pm 1.7 years), and had a smaller Cobb angle at diagnosis. Patients detected by screening had a higher chance of receiving brace treatment compared to otherwise detected patients (OR 3.1; 95% CI 1.3-7.0). In contrast to what would be expected, slightly more cases than controls were screened: 80.5% of cases were screened compared to 74% of controls (OR 1.44; 95% CI: 0.77-2.68; P=0.25). The authors conclude that screening for AIS does not reduce the need for surgical treatment for AIS and leads to more years of medical care and concern for the families.

12. There should be evidence that the complete screening programme (test, diagnostic procedures, treatment/intervention) is clinically, socially and ethically acceptable to health professionals and the public.

The discussion about the pros and cons of screening for AIS has been going on for years. ^{6,31} There are clear supporters and opponents of screening for AIS. Many expert opinions and individual studies support screening for AIS, which is in contrast with the majority of institutional recommendations and guidelines recommending against screening for AIS. ³¹

13. The benefit from the screening programme should outweigh the physical and psychological harm (caused by the test, diagnostic procedures and treatment).

Screening has not been shown to have beneficial effects compared to not screening.5 There are however wellestablished negative effects of screening. The test has many false positive results (see criterion 4). For adolescents with a false positive screening test there are adverse effects: concerns about having a (possible) physical defect and having medical/diagnostic examinations.^{2,32} The test has also many false negative (missed cases) results (see criterion 4). A false negative screening test may give a false sense of security, leading to delayed actions in cases where a scoliosis has been missed during screening or develops after screening. Adolescents diagnosed with mild scoliosis will have to be monitored for years to determine if progression will occur. If five percent of the patients with a Cobb angle of >10° will progress to >30°, 95% of these adolescents will be monitored without any consequences. 17,18

Pruijs et al. reported that the screening programme they studied tended to find non-progressive curves, while children with progressive curves were detected in other ways.³³

Brace treatment can have negative consequences on selfimage, peer relationships and quality of life, although varying effects have been described.^{34–37} A Cochrane review on the effectiveness of brace treatment described problems with heat under the brace, toileting and putting on and taking of the brace.²⁶

14. The opportunity cost of the screening programme (including testing, diagnosis and treatment, administration, training and quality assurance) should be economically balanced in relation to expenditure on medical care as a whole (ie. value for money). Assessment against this criterion should have regard to evidence from cost benefit and/or cost effectiveness analyses and have regard to the effective use of available resource.

In several studies the cost effectiveness of the programme has been evaluated. In a recent systematic review the cost effectiveness of school screening for scoliosis was investigated. Seven studies were included, no RCT's were found. The costs per screened adolescent varied from \$0.62 to \$61.03. The variation in qualifications and salaries of screeners, the type of screening, the criteria for diagnosis and the costs of monitoring and investigations explained the large variation in costs. The authors conclude that, based on current knowledge, no statement can be made on the cost effectiveness of screening for AIS.

Yawn et al. have calculated that the screening costs for finding one adolescent that needs (conservative or surgical) treatment are $$10.836.^{22}$

A recent Norwegian study calculated that screening costs \in 8.40 per child, \in 10,350 per patient braced, and \in 45,880 per patient operated. The authors make the assumption that screening leads to reduced treatment rates, and conclude that screening is cost saving when performed in girls only.³⁹

For the situation in the Netherlands, Bunge et al. state: 'If we assume that the OR of 0.64 is the true size of the effect, then the costs of keeping one patient from the need for surgery are estimated at (at least) 130,000 euros, and ± 5800 children would need to be screened. These are relatively high costs and involve considerable effort, given that severe scoliosis is neither common nor fatal. Furthermore, screening identifies some children who ultimately receive treatment but involves referral of many more who do not. Therefore, these costs are an underestimation of the real costs, because they exclude the costs of visits to general practitioners and orthopaedic surgeons and of radiographs attributable to false-positive results.'⁵

15. All other options for managing the condition should have been considered (e.g. improving treatment, providing other services), to ensure that no more cost effective intervention could be introduced or current interventions increased within the resources available.

Based on the current search, we cannot answer the question if this criterion is met.

16. There should be a plan for managing and monitoring the screening programme and an agreed set of quality assurance standards. The described literature search did not reveal any literature on this aspect. In an expert opinion article pleading in favour of screening for AIS, several suggestions are made to optimise a screening programme for AIS.⁶

17. Adequate staffing and facilities for testing, diagnosis, treatment and programme management should be available prior to the commencement of the screening programme.

Based on the current search, we cannot answer the question if this criterion is met.

18. Evidence-based information, explaining the consequences of testing, investigation and treatment, should be made available to potential participants to assist them in making an informed choice.

The described literature search did not reveal any literature on this aspect.

19. Public pressure for widening the eligibility criteria for reducing the screening interval, and for increasing the sensitivity of the testing process, should be anticipated. Decisions about these parameters should be scientifically justifiable to the public.

At this moment, evidence based information is missing on the proper frequency and age for screening for scoliosis.

Discussion

Screening for AIS aims at detecting patients in an early stage of the clinical course, in order to apply conservative treatment to try and prevent further progression of the curve and the need for surgical treatment. For a screening programme to be effective, several criteria have to be met. These criteria are described by the UKNSC, divided into four aspects: the condition, the test, the treatment and the screening programme. We used the available (recent) literature to investigate if the criteria for a screening programme for AIS are met, focussing on YHC in the Netherlands.

Regarding the condition, we conclude that although mild AIS curvatures are relatively frequent the clinically relevant curves have a low incidence. The natural history of AIS is not predictable. This means that, if scoliosis is diagnosed, many adolescents often will have to be monitored to determine if progression will occur. It cannot be predicted which adolescents will need brace treatment in the future. The period of monitoring can take several years, and can give rise to concern for adolescents and their parents and leads to health care costs. Without a screening programme, at least some of the adolescents with a (progressive) scoliosis will be detected in other ways (such as a parent or gym teacher). The optimal age and frequency of screening are not known.

Regarding the test, we conclude that the FBT is not valid enough for use in a screening programme. The PPV of 2.6% and the sensitivity of 56% for (conservative or surgical) treatment mean that a considerable number of adolescents will be unnecessarily referred for further investigations and that a considerable number of patients will be missed.

Regarding the treatment, we conclude that there seems to be sufficient evidence to support brace treatment for preventing progression of AIS. There is no high quality evidence to recommend the use of SSE for AIS.

Regarding the screening programme, we conclude that there are no RCT's on the effectiveness of screening for AIS. A case-control study could not detect a beneficial effect of screening for AIS. The ongoing debate about screening for AIS implies that a screening programme is not acceptable to all health professionals. There is no evidence to support that screening has more beneficial effects than not screening. This can be due to the low validity of the screening test and the unpredictable natural history. There are clear disadvantages to the screening programme due to the high number of false positives and false negatives. It is not clear if the screening programme is cost effective. In a recent study, Adobor et al. conclude that screening can be cost effective, assuming that screening leads to reduced treatment rates.³⁹ However, based on the literature, we consider this assumption questionable.5

Many expert opinions and individual studies support screening for AIS, which is in contrast with the majority of institutional recommendations and guidelines recommending against screening for AIS.³¹ Perhaps this contrast is caused by the different viewing points of clinicians and public health specialists. A clinician may approach the problem mainly from the patient point of view and wants to prevent adolescents from needing surgical therapy. The public health specialist approaches the problem from a population point of view and tries to assess whether the benefits of screening outweighs the harm of screening.

The recent successful brace treatment RCT for AIS cannot compensate the other screening problems. Therefore, we conclude that there is not enough evidence to support a screening programme for AIS.

At this moment, even the diagnostic methods available in a clinical setting cannot reliably predict which patients with a Cobb angle $\geq\!10^\circ$ will have a progressive scoliosis needing treatment and which patients with a Cobb angle $\geq\!10^\circ$ will have a non-progressive curve. A method is needed that can make a reliable distinction between these two groups. If this method is also suitable in a screening setting, population based screening may become a viable option.

Author statements

Ethical approval

Not required (this study is based on available literature).

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Competing interests

None declared.

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