UK National Screening Committee

Screening for Adolescent Idiopathic Scoliosis

External review against programme appraisal criteria for the UK National Screening Committee (UK NSC)

Version: 3

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The UK NSC advises Ministers and the NHS in all four UK countries about all aspects of screening policy. Its policies are reviewed on a 3 yearly cycle. Current policies can be found in the policy database at http://www.screening.nhs.uk/policies and the policy review process is described in detail at http://www.screening.nhs.uk/policyreview

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Introduction

The UK National Screening Committee does not currently recommend screening for adolescent idiopathic scoliosis. The committee cited a report published by the US Preventative Services Task Force from 2004 as evidence supporting the policy. The policy was reviewed in 2006 but no significant changes were made. For this review we have assessed publications relating to adolescent idiopathic scoliosis (AIS) published since the end of the systematic search for the USPSTF review (2002).

The US Preventative Services Task Force review and a summary of recommendations are available from http://www.uspreventiveservicestaskforce.org/uspstf/uspsaisc.htm

The key points which led to the recommendation were:

- Screening asymptomatic adolescents did not detect important degrees of idiopathic scoliosis at an earlier stage than detection without screening
- The accuracy of the most common screening test, the forward bend test with or without a scoliometer, in identifying adolescents with idiopathic scoliosis is variable, and there is evidence of poor follow up of adolescents with idiopathic scoliosis who are identified in community screening programmes
- Treatment of idiopathic scoliosis during adolescence leads to health benefits (decreased pain and disability) in only a small proportion of people
- Most cases detected through screening will not progress to a clinically important forms of scoliosis
- Scoliosis needing aggressive treatment, such as surgery, is likely to be detected without screening
- Treatment of adolescents with idiopathic scoliosis detected with screening may lead to moderate harms, including unnecessary referral for speciality care

Given these findings, the USPSTF concluded that the harms of screening adolescents for idiopathic scoliosis exceeded the potential benefits.

At the time of the NSC 2004 report there was a limited evidence base for the effectiveness of screening tests and programmes for adolescent idiopathic scoliosis and for the effectiveness of early conservative interventions such as exercise programmes or brace treatments. The major limitation of the evidence was a lack of randomised controlled or prospective controlled studies. The available evidence came from lower quality, often non-comparative, studies including people who had not been identified through screening. The studies of the standard scoliosis screening techniques, used prior to radiography, found a poor positive predictive value for detecting clinically significant scoliosis (based on Cobb angle measurement) and did not reliably identify individuals who would require treatment. At the time of the USPSTF review there had not been an RCT or controlled trial of a screening programme for scoliosis.

The USPSTF report has reviewed journal articles from 1994 to 2002. An updated systematic search has been performed for and based on this the studies published from 2002 to 2011 have been assessed. We have prioritised study designs such as systematic reviews of comparative studies, randomised controlled trials (RCTs) and large prospective controlled trials for inclusion in this review. Where relevant we have presented the evidence referred to in the USPSTF review as a base line for the assessment of new evidence against the UK NSC criteria.

Appraisal against UK NSC Criteria

These criteria are available online at http://www.screening.nhs.uk/criteria.

1. The condition should be an important health problem

Adolescent idiopathic scoliosis (AIS) is a three dimensional curvature of the spine that presents during puberty in healthy children and is not related to an underlying pathology. The degree of curvature is the marker for scoliosis and is generally measured by Cobb angle on a radiograph taken from the coronal plane view; although curves in the lateral plane also impact on the evolution of scoliosis[1]. A Cobb angle of over 10º is generally considered clinically significant as more minor curves than this have little chance of progressing. Adolescent idiopathic scoliosis can either improve, remain stable or progress over adolescence, the natural history is influenced by the degree of curvature, age and skeletal maturity, type of curve at presentation and for females, menarchal status.[2]

The prevalence of adolescent idiopathic scoliosis is estimated 2% to 3% of children between 10 and 16 years of age, using a definition of over 10º spine curvature.[3] Larger curves present at a lower frequency and it is estimated that 40 degree curves make up 0.1% of the total AIS population.[4] Whereas the frequency of curves between 20 and 30 degrees is approximately 0.3 to 0.5%.[4] A recent Japanese cross-sectional study assessed the prevalence of curvature over 10º in an 11 to 12 year old age group and a 13 to 14 year old age group. For girls the prevalence was 0.78% in 11 to 12 year olds and 2.51% in 13 to 14 year olds. In boys the prevalence was 0.04% in 11 to 12 year olds and 0.25% in 13 to 14 year olds.[5] A Korean study found that the prevalence rate in girls aged 10 to 12 was 5.57%, and 3.90% in 13 to 14 year old girls. The study found the prevalence was 2.37% in 10 to 12 year old boys and 1.42% in 13 to 14 year old boys. No prevalence studies for a Western European adolescent population were retrieved from the updated search.

The prevalence of adolescent idiopathic scoliosis is greater in females than males. In cases where the curvature is around 10 degrees the male to female ratio is around 1:1, when the disorder is over 30 degrees the ratio changes can rise to 10:1 in favour of females.[4]

What impact does scoliosis have on health and quality of life?

The U.S. Preventative Services Task Force (USPSTF) review used data from two retrospective cohort studies. The first presented data from a questionnaire completed by a retrospective cohort that included 34 patients with adolescent idiopathic scoliosis and 31 age- and sex-matched controls. They had an average follow up of 22 years at which time the patients were on average 36 years. The research found that 65% of the AIS patients and 32% of the control group reported back pain. Nine per cent of the scoliosis group said that they had severe back pain compared to no one in the control group. The USPSTF had reported from this study that perception of disability was comparable in both groups but noted that no scores or p values had been given in the study.

The second assessed 1,476 clinic-based patients more than 10 years after referral. The patients were reported to have Cobb angles <20 to >40 degrees in the USPSTF review. Compared to a control population, who were age and sex matched, back pain in the past year was higher in the study group (OR 2.14; 95% CI 1.81 to 2.53). Female patients reported a poorer overall perception of health than women in the control group (OR 1.19; 95% confidence interval [CI] 1.06 to 1.34). Women with idiopathic scoliosis during adolescence also showed significantly greater difficultly in performing physical activities than did women in the control group.

In the updated search further primary studies were identified that had compared back pain, disability or quality of life in people with scoliosis compared to people without scoliosis. One Japanese cross-sectional study[6] suggested higher reporting of back pain in adolescents that had been screened positive for scoliosis (during the first stage of a three step screening process) compared to those screened negative, However, in this study diagnosis of scoliosis had not been confirmed.

One study [7] in the updated search had compared the health related quality of life of 226 German female patients with idiopathic scoliosis to that of age- matched population norms. The study was conducted between May 1998 and February 1999 and had recruited inpatient volunteers, although it should be noted that these patients may not be representative of the whole female IS population. The study used three QOL tools: patients aged 17 years or older were assessed with the SF-36, children and adolescents were assessed with Berner Questionnaire for Well-Being (BFW) and the emotional state of the children and adolescents was measured with the State-Trait-Anxiety-Inventory for Children (STAIC). German language versions of these tools were used. German population norms including subgroups of people with self-reported back pain were used as the comparator for the SF-36. Data from a cohort of Swiss children aged between 14 and 20 years was used as a comparator for the BFW and comparative values from 217 German school children with a mean age of 11 years was used for the STAIC. There were 146 adolescents aged between 11 and 16 years of age, 36 participants aged between 17 and 21 years and 44 individuals aged over 21 years. The average Cobb angles were 31.8^{o} (SD 14.63) in the 11-16 years group, 28.79^{o} (SD 11.55) in the 17-21 years group, and 47.85^{o} (SD 21.21) in the over 21 years group. A large proportion (60.3%) of the 11 to 16 group were wearing braces at the time of the study, 36.1% of the 17 to 21 group were wearing a brace, whereas no one over 21 was wearing a brace.

The study found that adolescents with AIS aged between 11 and 16 had less of a positive attitude to life, more body complaints and reactions, lower self-esteem and greater depressed mood, but enjoyed positive events in their life more than the population norm. However, as 60% of the adolescents were wearing braces, these quality of life scores may reflect a combination of the impact of the disease and of the treatment. The adult population reported more psychological and physical impairment than the population norm.

A small Japanese study[8] evaluated untreated Japanese patients with idiopathic scoliosis using the Scoliosis Research Society Outcomes instrument (SRS-24) and compared these patients to a non-scoliosis group. There were 141 patients surveyed with a mean age of 13.6 years (range 10-17 years) who had idiopathic scoliosis that was not treated and had a Cobb angle of over 20^{o}. Seventy two healthy junior high school students were also surveyed using the SRS-24. The Idiopathic scoliosis group were classified as having a mild deformity if their Cobb angle was less than 30^{o}, moderate deformity if the curve was measured between 30 and 49^{o} and severe deformity if the Cobb angle was more than 50^{o}. They found that patients in the severe deformity group had the lowest scores in pain and self-image compared to the other scoliosis groups and the non-scoliosis group. The scoliosis group had lower self-image about their back appearance compared to the non-scoliosis group but for general self-image had higher scores. The researchers highlighted that baseline scores in a Japanese population using the SRS-24 scores may differ from the western population because of cultural differences and that the internal consistency using Cronbach's alpha for all domains was considerably low.

Long term clinical outcomes

The USPSTF review did not report an increased mortality rate. In the updated search a systematic review[4] stated that untreated AIS does not increase the mortality rate overall, but they report that on rare occasions it can progress to greater than 100 Cobb degrees and in some of these severe cases it may increase likelihood of mortality In the updated search we found no further primary studies that had compared long term mortality outcomes for individuals with adolescent idiopathic scoliosis compared to a control population.

A Cochrane review [9] of the use of braces for scoliosis discussed in its narrative introduction that if scoliosis surpasses a critical threshold, usually considered to by Cobb 30^{o} at the end of growth that the risk of health problems in adulthood increases. They said that problems include reduced quality of life, disability, pain, increased cosmetic deformity, functional limitations, sometimes pulmonary problems and possible progression during adulthood. The authors of this review said that because of this, management of scoliosis also involves the prevention of secondary problems associated with the deformity.

Other than quality of life since the USPSTF review there have been no primary studies that have assessed clinical outcomes in people who have AIS compared to people without scoliosis over the long term.

The importance of AIS as a health problem remains unchanged since the USPSTF review in 2004.

Criterion 1 met? Yes. The condition is an important health problem. Adolescent idiopathic scoliosis is associated with poorer quality of life and pain in some people, however, the degree of curvature and the impact on quality of life may vary between individuals.

2. 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.

It is estimated that only 10% of adolescents diagnosed with scoliosis have curve progression requiring medical intervention and more than 90% of diagnosed cases require only observation with repeated examination during the growing years.[4] Curves can have a thoracic, lumbar, thoracolumbar, or a double curve pattern. These curve patterns may have their own particular characteristics and predicted course.[10] The USPSTF review in 2004 did not address the natural history of curve progression in their review. However, knowing whether a curve is likely to progress is of importance in determining whether conservative treatments are indicated as the aim of some of these treatments is to halt progression rather than correct existing curves. It is also important to note that a proportion of individuals with AIS will show improvement as they continue to grow.

Outside of the updated search we found a paper from the 1980's[10] which discussed the natural history found in a cohort of individuals with adolescent idiopathic scoliosis. Out of a cohort of around 100 people that had not received treatment for AIS, 68% of curves progressed greater than 5% after skeletal maturity. The progression depended on the type of curvature and the severity of curvature, for example curves over 30^{o} tended to show a greater rate of progression. However the author noted that with reference to curvature progression there are probably many natural histories and treatment decisions must be individualised.

Predicting progression

A second older natural history study followed a large cohort of 727 patients with untreated scoliosis during growth between 1970 and 1979.[2] The cohort study defined progression dependent on the initial curve. For an initial curve of 19^{o} or less progression was defined as an increase of at least 10^{o}, with a final curve of over 20^{o}. For an initial curve of between 20 and 29^{o} progression was defined as an increase of 5^{o} or more. They found that 23% of the population showed curve progression and 11% showed curve improvement (by 5 degrees or more) between the initial visit and the final follow up (when the adolescents were skeletally mature or until progression of the curve had occurred). The authors determined that the factors which had the greatest correlation progression were the magnitude of the curve, Risser sign and the patients' chronological age.

A progression factor could be estimated from the following formula:

PF= (Cobb angle -3 x Risser Sign)/Chronological age

The Society for Scoliosis Orthopaedic and Rehabilitation Treatment (SOSORT) produced guidelines that recommend treatment options depending on this progression factor.[11] Whereas other approaches use Cobb angle cut offs for treatment (with braces) with these adjusted for people with smaller Cobb angles but a greater progression factor .[4]

In the Lonstein and Carlson paper [2] the authors had highlighted the difficulties they had in producing a prediction model. They confirm that with the data available it is impossible to predict with total accuracy which curve will progress and which will not. It is possible to determine the likelihood of progression at the initial examination, but in only a very general manner. The updated search did not identify a study that had determined the sensitivity of this progression factor using defined cut-offs in another cohort.

In the updated search we identified one study conducted in Singapore in which adolescents with scoliosis (Cobb angle ≥ 10^{o}) who had been identified through a school screening programme were followed up until skeletal maturity. The researchers said that as previous studies had suggested that curves under 30^{o} were unlikely to progress after skeletal maturity, they aimed to identify the prognostic factors for curve progression to a magnitude of 30^{o} at skeletal maturity, in skeletally immature patients with AIS. Two hundred and sixty two children out of 72,699 children aged six to nine years who had been screened were followed. Of the 262 adolescents with scoliosis, 226 were skeletally immature (Skeletal maturity was defined as an age of 15 years or a Risser grade of 4 or 5). These patients were followed and assessed at three to six month intervals until they were skeletally mature. One hundred and eighty six patients had complete follow up and 20.4% (38 adolescents) progressed to a Cobb angle of 30^{o} or more at skeletal maturity. Receiver operator curve analysis showed that Cobb angle at presentation was the most important factor in determining which curves would progress. An angle of 25^{o} at presentation was associated with a positive predictive value of 68.4%, a negative predictive value of 91.9% and the ROC value was 0.80 for a curve of 30^{o} at skeletal maturity.

Three additional studies had assessed additional risk factors or prognostic factors for progression of scoliosis, other than Cobb angle, bone maturity or age, were identified in the updated search. As progression risk was not described in detail in the USPSTF evidence review, these can be considered as new for this update. However, it should be noted that studies assessing the risk of progression published outside of the search dates of the current update search (i.e. before 2002), were not systematically reviewed for this update.

Rate of growth

Ylikoski 2005[12] published a retrospective analysis of records from 535 consecutive girls with untreated AIS from one orthopaedic hospital in Finland and found that the rate of progression was most notable during the growth spurt; there was a trend for increased progression velocity with increased growth velocity. For example patients growing at a rate of less than 1 cm per year had on average a progression velocity of their major + minor curves of 1.6°/year, patients with a growth velocity of 2 cm/year had a progression velocity of 5° a year and those with a growth velocity of ≥ 6 cm/year had an average major and minor curve progression velocity of 9.0°/year.

Osteopenia

Hung et al 2005[13], suggested that 27% to 38% of girls with adolescent idiopathic scoliosis have systemic osteopenia. The study measured bone mineral density in Chinese girls with and without AIS and generated an age adjusted bone mineral density z-score. An age-adjusted bone mineral density z-score of less than or equal to -1 was classified as osteopenia. The study also assessed whether osteopenia was a prognostic factor for in predicting curve progression. They performed a prospective study which followed 324 girls with AIS, 27.5% and 23.1% of this cohort had osteopenia at the spine and hips respectively. The girls were on average thirteen and a half years of age. A larger initial Cobb angle (curve ≥ 40°, odds ratio [OR] 4.6, 95% CI 1.3 to 15.9), a lower Risser grade (OR 4.7, 95% CI 2.2 to 9.9) , premenarchal status (OR 2.5, 95% CI 1.0 to 6.0), a younger age at diagnosis (OR 2.1, 95% CI 1.1 to 4.0) and osteopenia in the femoral neck of the hip on the side of concavity (OR 2.3, 95% CI 1.1 to 4.5) were found to be predictive of curve progression. Using these risk factors a predictive model was produced. The area under the receiver operating characteristic curve of the model was 0.80 ($p<0.01$).

DNA marker panel

A DNA marker panel of 53 SNPs was assessed in three cohorts with known outcomes to determine clinically meaningful cut-offs in determining risk of severe curve progression. Progression to a severe curve was defined as progression to a >40° curve in an individual still growing, or progression to a >50° curve in an adult. Prior to the DNA tests, using the participant records, the age and Cobb angle at presentation was plotted against risk of progression to a severe curve. A low-risk patient group was defined as those people who had a <1% risk of progression. Multiple logistic regression on the 53 DNA markers was used to generate a AIS-prognostic test score. A low risk score had a cut off of less than 41. The cohorts included 277 low-risk females representing a screening cohort, 257 females representing higher risk patients followed at referral centres and 163 high risk males. Low risk scores had negative predictive values of between 97% and 100% in the three cohorts. Using a cut-off score of 50 and 180 could identify 75% of people who were at low risk of curve progression with a less than 1% chance of requiring surgical intervention for their scoliosis. The researchers had highlighted that their DNA marker panel test only applied to white populations and the test had not been optimised for people of different ethnicities.[14]

Criterion 2 met? Not met. The prevalence of AIS and of the prevalence of curvature of different severities has been estimated in various cohorts. Some analysis of natural history has been performed to determine risk factors for progression, which is important in order to determine which patients are indicated for treatment to prevent progression. However, further validation is required of the effectiveness of risk prediction models. These are not sufficiently accurate for routine use.

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

We identified no studies, in the updated search that assessed potential primary prevention interventions. As adolescent idiopathic scoliosis is without known cause and it is unclear when the age of onset is, we felt that this criterion was not applicable.

Criterion 3 met? Not applicable.

4. If the carriers of a mutation are identified as a result of screening the natural history of people with this status should be understood, including the psychological implications.

Not applicable.

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

There are various non-invasive tests that assess for curvature prior to referral for radiographic examination to measure the extent of the curve and skeletal maturity. The screening tests are simple and safe (they are non-invasive) but lead to additional radiography follow up. It has been suggested in a narrative review endorsed by the American Academy of Orthopaedic surgeons, the Scoliosis Research Society, the Pediatric Orthopaedic Society of North America and the American Academy of Pediatrics that adolescents without significant spinal deformity who are referred to a specialist for evaluation often do not require radiographs. They note that current radiography techniques have lowered the exposure to radiation compared to the past.[15]

The 2004 USPSTF review described, primarily, screening programmes carried out in schools. The non-invasive tests used in this setting require referral for further evaluation. The test accuracy was calculated for these initial screening tests based on either the Cobb angle or the rates of adolescents who require treatment for their scoliosis as the gold standard. It was not described in this review how the adolescents were managed after referral or how many adolescents had radiographs or not. Without this information, the safety risks of these screening programmes cannot be fully determined. Increased referral for radiography is discussed in Criterion 13.

Tests commonly used in screening programmes include:-

The **Adam's forward bend test:** this requires that the child bend over at their waist as if to touch their toes. The examiner, with eyes level at the back looks for one side being higher than the other. The Italian Guidelines[1] suggest that the test's positive predictive value varies since it is proportional to the degree of curvature and depends on the examiners experience and the degree of curvature. The forward bend test is cited as having a relatively low specificity of 77.8%, leading to the referral of a significant proportion of false positives.[16]

The Italian guidelines[1] suggested that to improve intra-examiner reproducibility a scoliometer may be used. The scoliometer measures the angle of trunk inclination (ATI) and permits the determination of cut-off points above which radiographic study is indicated. Citing studies published before 2004, the guidelines said that this tool has a sensitivity of about 100% and a specificity of about 47% when an ATI angle of 5^{o} is chosen and 83% sensitivity, 86% specificity when using an ATI angle of 7^{o} as a cut-off.

The **hump-meter** (level protractor) measures the height of the difference between curve concavity and convexity and the Italian Guidelines suggest that a cut-off point of 5 mm should be used to measure back hump.[1]

Moiré topography: produces a contour map of a person's back where asymmetry of the contour lines are assessed. Contour lines are mapped by shining light through a screen of parallel strings which casts shadow "fringe patterns" dependent on the topography of the back (when the person is standing upright). The contours form different characteristic patterns for convex and concave sections of the back. The angle-from-horizontal between corresponding contour lines measured on the left and right of the back are measured.

These tests lead to referral where scoliosis is confirmed and measured with a radiograph to determine Cobb angle. The measurement of Cobb angle from radiographic film has been cited as having intra- and inter-observer variability of 3 to 5^{o} and 6 to 7^{o} respectively.[1] We found two studies that discussed alternative curve measurement techniques to Cobb angle. [17,18] It is unclear if these alternatives are widely used.

Additional small studies were identified in the updated search that had assessed a dynamic balance test and a cell based assay measuring melatonin signalling.[19] However the findings from these preliminary small studies require validation in prospective cohorts.

What is the positive predictive value of screening for scoliosis?

The USPSTF update described two cohort studies (one retrospective, one prospective) that assessed referral rates following screening for further radiological assessment and the proportion that required treatment. The first US study found that the incidence of scoliosis that required treatment in the screened population of 2042 children (screened annually from grade 5 (around age ten) to grade 19 (around 14 years old) and followed until age 19 was 0.4% and the positive predictive value of the scoliosis programme was 0.05 (95% CI 0.048 to 0.052). The second prospective cohort study in Greece screened 21,220 children aged 8 to 12 years. Of these children 9.6% (over 2000) were referred for further radiological treatment and 0.06% of the cohort (13 children) required treatment, having Cobb angles over 20 degrees. It should be noted that the prevalence of scoliosis requiring treatment in these cohorts is likely to be influenced by the age range of children that were screened. The USPSTF review did not detail the number of children who were not referred for radiography but did go on to develop a clinically significant scoliosis requiring treatment (the false negative rate) or the sensitivity of screening.

Since the publication of the USPSTF update, a systematic review of the clinical effectiveness of school screening programmes has been published.[20] This SR searched for retrospective cohort studies published up to 2008. Studies were included if they considered a screening programme that used either the forward bending test (FBT), angle of trunk rotation (ATR), or Moiré topography for adolescents only. Other inclusion criteria included: studies that reported the results of screening tests and radiographic assessments, reported the incidence of curves with a minimum Cobb angle of 10^{o} or greater, and reported the number of referrals for radiography. Thirty six studies from 17 countries were included, with publication dates ranging from 1977 to 2005. The screening programmes/studies used different screening tests. Twenty three (64%) studies used the FBT as the only screening test. Eight (22%) studies additionally measured the ATR, and 2 (6%) further used Moiré topography. One other study used the FBT and Moiré topography and 2 others used Moiré topography and low-dose X-ray. There was great heterogeneity between the studies with an I^2 of over 90%. Use of the FBT alone resulted in a higher referral rate (7.2% vs. 2.6%) and lower PPV for curves ≥ 10 degrees (23.2% vs. 38.0%) and ≥20 degrees (3.5% vs. 11.0%) than combination screening tests. However, there was no evidence that the use of FBT alone influenced the detection of students requiring treatment and only 13 of the 36 studies reported treatment outcomes. The one study that calculated sensitivity

screened 2242 children and had been reported in the USPSTF update.[21] The study had follow-up information from screened students through skeletal maturity. The sensitivity was 64% (95% CI 45.2% to 82.8%) for curves ≥20 degrees and 55.6% (95% CI 23.1% to 88.0%) for treatment. This means that approximately 44% of people in this cohort who received treatment for scoliosis were not identified through the screening programme.

Pooled estimates from the systematic review of school screening are summarised in Table 1 below. The author's conclusions of this review were that "there was substantial heterogeneity across studies due to the use of different screening tests and different study sizes. The use of FBT alone in school scoliosis screening is insufficient. To properly assess the clinical effectiveness of school scoliosis screening we need large retrospective cohort studies with students followed by skeletal maturity. This assessment could be facilitated by the continuation of school scoliosis screening programmes."

Table 1: A pooled analysis of the effectiveness of school screening programmes. Reported in Fong et al 2010[20]

Outcome	Number of Studies	Pooled estimates across the studies	Heterogeneity (I^2)
Prevalence Cobb angle ≥ 10 degrees	34	1.3% (95% CI 1.0% to 1.7%)	98.9%
Prevalence Cobb angle ≥ 20 degrees	28	0.2% (95% CI 0.2% to 0.3%)	97.6%
Prevalence Treatment	13	0.2% (95% CI 0.0% to 0.3%)	96.1%
Referral rate	36	5.0% (95% CI 3.3% to 6.7%)	99.9%
Positive Predictive Value Cobb angle ≥ 10 degrees	34	28.0% (95% CI 21.3% to 34.7%)	98.9%
Positive Predictive Value Cobb angle ≥ 20 degrees	28	5.6% (95% CI 2.9% to 8.3%)	98.9%
Positive Predictive Value Treatment	13	2.6% (95% CI 0.9% to 4.2%)	94.5%

This systematic review[20] had included retrospective cohort studies. Additional studies were identified in the updated search that had assessed screening programmes that had not been included in this review.

Since the publication of this systematic review two large studies, one prospective cohort (including retrospective analysis) and one cross-sectional study have assessed screening programmes for AIS.

The high-referral and false positives rate of school screening for scoliosis has led to suggested recommendations on how to improve the clinical effectiveness of school screening by (1)

selecting populations eligible for screening who were at higher risk of developing clinically significant AIS and (2) using more objective referral criteria than the forward bend test (Bunnell et al 2005, cited in Lee et al 2010.[22]) Suggestions for improving the objectivity of referrals were to use a scoliometer to measure the angle of trunk rotation (ATR) during a forward bend test, applying specific ATR cut-offs for referral[22] or to use Moiré topography. However, as this Moiré topography is relatively more expensive it is recommended that it is used as a second tier test.[22]

The first study had two publications[22,23] which had assessed a two tier screening programme using ATR angle and Moiré line cut-offs in a Hong Kong cohort of 115 and 178. The aim of one of the analyses[22] was to assess the key determinants of the referral rate for radiography, positive predictive value, negative predictive value, sensitivity and specificity. Adolescents with clinically significant scoliosis were defined as individuals with curvature of at least 20^{o} Cobb angles by the age of 19 years.

This Hong Kong screening programme was a biennial voluntary screening programme for students aged 10, 12 and 14 years. The students were screened with the forward bend test and their ATR measured. If the ATR was 3^{o} or 4^{o} the students were rescreened more frequently. If their ATR was between 5^{o} and 14^{o}, or if students displayed any obvious clinical signs of back or shoulder asymmetry then they were tested using the second tier of screening by ATR and Moiré topography. Following this second tier screening, individuals who had ATR of 15^{o} or above, two or more Moiré lines, or did not meet these two criteria but were found to have significant clinical signs of scoliosis, including uneven shoulder height, pelvic tilt, rib or loin hump, scapular prominence, and/or truncal shift, were referred for radiography.

The study found that 6.4% of the participants underwent second tier screening. Out of the whole cohort, 2.1% were referred for radiography (based just on objective measures, 2.8% if observations based on clinical signs were also included). Clinically significant scoliosis (Cobb angle ≥ 20^{o}) was observed in 1406 students (1.2% of whole cohort) referred for radiography by screening and 190 (0.16%) who were referred by other sources. Using the protocol referral criteria; ATR, Moiré topography or the presence of significant clinical signs, the PPV for clinically significant scoliosis was 43.6 % (95% CI 41.8% to 45.3%). The negative predictive value was 99.9% (95% CI 99.8% to 99.9%), the sensitivity was 88.1% (95% CI 86.4% to 89.6%) and the specificity was 98.4% (95% CI 98.3% to 98.5%). The researchers reanalysed their data to determine what the effect of screening by just one gender or age group would be. They determined that a strategy of screening girls only would be inappropriate, as it would miss 18.3% of children with ≥ 20^{o} curvature. As not many students had curvature of ≥ 20^{o} detected at the age of 10 years, the study concluded that screening before the age of 10 may not be necessary. They further suggested that boys could begin screening after 12 years of age.

The study also looked at the effect of using different combinations of screening tests. Based on their analysis the researchers recommended the inclusion of objective screening tools as well as clinical signs. Without the use of Moiré topography, sensitivity dropped to 39.8%. Clinical signs were also important, without their use the sensitivity dropped to about 55.5%. The benefit of clinical observation is expected to be dependent on the experience of the screeners. A screening programme recruiting non-clinical workers as screeners would require that these people had received sufficient training from an expert. It should be noted that this cohort included mainly Chinese adolescents who may show different growth patterns to other ethnic groups.

The other publication that had described this screening cohort[23] assessed the effectiveness of the school screening programme. In addition to the 115,190 individuals who went through school screening, there were 42,203 students who had chosen not to go through screening. Of

these non-screened adolescents 71 (0.17%) had AIS detected by the age of 19 (Cobb angle over 10^{o}). This proportion (0.17%) represents the number who would be expected to present without screening.

Of the 115,190 students who went through the screening programme, 2619 (2.2%) had AIS detected. Of the students who went through screening 97% (111,950) of students were not referred for radiography. Of these 111,950 were not referred for radiography through the screening programme, 271 went on to have radiography outside of the programme. Of these 246 had AIS detected by the age of 19 (false negatives).[23] This gave an overall prevalence of AIS in the screened cohort of about 2.5%. This was substantially higher than the proportion of individuals presenting with AIS in the unscreened population (0.17%), suggesting that the majority of adolescents with AIS do not present without screening.

The second study was a prospective observational study of a screening programme to determine the trends in prevalence of AIS in the Korean population.[24] The study reported that the prevalence of scoliosis increased progressively from 1.66 to 6.17% between 2000 and 2008. In their discussion the researchers noted that the number of children screened each year varied and this may have confounded the prevalence estimates. For example, in 2000, 8,780 boys and girls were screened compared to 74,701 in 2008. The screening programme screened students in two provinces in Korea who were aged between 10 to 14 year old, the screening was performed by an experienced team of nurses who used the forward bend test with a scoliometer. Individuals with an ATR of over 5^{o} were referred for radiography. Cobb angles of $\geq 10^{o}$ were considered to be clinically significant. The cumulative PPV over the 8 years was 46.4%. They noted that there was a trend for the PPV to be lower for boys than girls 41% vs. 51%.

Neither of these studies described the PPV of screening for predicting individuals who required treatment, or the proportions of adolescents identified through screening who had successful subsequent treatment (i.e. with no further progression). However, they did have better PPV for detecting clinically relevant curvature (defined as $\geq 20^{o}$ in the Hong Kong study and $\geq 10^{o}$ in the Korean study) than the screening studies reported in the USPSTF review and those included in the Fong et al 2010 systematic review.

There was an additional study in the updated search [25] that had compared referral rates for radiography following two screening approaches. The first approach involved a school physician performing the initial clinical investigation followed by uncertain cases referred to an orthopaedic surgeon. The second approach involved assessment by an orthopaedic surgeon only. In the study 5,731 children aged 9 to 14 years were assessed by the first two-step approach and 3,264 were assessed by the second approach. The study found that 1.5% of the first approach group and 2.91% of the second approach group had X-ray examinations performed. However this study did not detail the specificity of these approaches, i.e. it was not clear what proportion of the students that had an X-ray had been demonstrated to have clinically significant scoliosis.

Criterion 5 met? No. There still remains uncertainty about the precision or diagnostic accuracy of the screening tests. The positive predictive values for any test identifying clinically significant scoliosis according to Cobb angle or those who need treatment are low and vary between studies. This means that there is a risk that screening will lead to a high number of false positives in a low prevalence setting. These people may receive unnecessary follow up or treatments which is a potential harm of the screening programme. The screening programmes also vary in the age range over which screening takes place and the number and frequency of screening

tests that a person receives over their adolescence and there is limited data on the sensitivity of screening tests at these different ages and frequencies to allow. No validation studies were identified.

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

A Cobb angle of over 10^{o} is considered to be clinically significant by the Scoliosis Research Society. Cobb angle cut-off for treatment is discussed in Section 10. For the screening tests, the forward bend test is a non-quantitative test but a scoliometer can be used to measure the angle of trunk inclination (ATI) or angle of trunk rotation (ATR). Quantitative measures of back surface asymmetries can be obtained using Moiré topography.

The updated search identified no evidence-based guideline cut-off levels for screening tests following systematic review. The Italian guidelines made a non-consensus opinion recommendation that a scoliometer/ATI cut-off point of 5^{o} or a 5mm back hump could be used for referral for radiography. There was one survey which had been completed at the 4th International Conference on Conservative Management of Spinal Deformities.[26] The aim of the survey was to determine whether there was consensus on current screening practices from 35 representatives from thirteen countries including 4 from the UK, 5 from Israel, 9 from North America, 1 from Japan and 16 from other European countries. Six of these were orthopaedic surgeons, two were orthopaedic surgeons and physiatrists (rehabilitation medicine specialist doctors), five were rehabilitation doctors, two were physiotherapists and orthopaedic surgeons, nine were physiotherapists, one was a nurse, and the remaining others were health care providers from other disciplines. Approximately a quarter performed a school scoliosis programme at their centre. Twenty per cent said that a school screening programme had been discontinued at their place of work.

The participants were asked *"Over which ATI or ATR is a hospital consultation and/or radiographical examination recommended?"* The survey sample did not reach a consensus. One person reported more than 4 degrees of ATI/ATR, 10 people reported more than 5 degrees, 8 people reported more than 6 degrees, 2 people reported more than 7 degrees and 1 person reported more than 8 degrees. 13 people said that scoliometer measurements were not sufficient to decide or gave no information for this question.

We found no systematic reviews or surveys on appropriate Moiré topography outcomes which would be indicate to referral for radiography.

Criterion 6 met? No. There is no clear consensus or evidence-based recommendation on a suitable cut-off level for screening tests which could be used as a threshold for radiography follow up.

7. The test should be acceptable to the population

The survey described further in Criterion 6, asked "*Has your centre encountered non-cooperation or refusal of the screening examination from children or their parents*?" Three responders said yes and 15 said no. Seventeen people gave no information. In their discussion they said "*The responders reported that 2% refused school screening. This should be considered an acceptable rate, as many children are present at the following years screening. It was also reported that the encountered difficulties in performing the school screening were not usually*

the non-cooperation or refusal of the screening examination by parents but mainly the negative attitude from mainly older children".[26]

We found no primary studies that had assessed parent and adolescent acceptability of the non-invasive screening tests or radiography follow up.

Criterion 7 met? No. It is unclear whether the test would be acceptable to the UK population.

8. 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

The discussion surrounding the survey which was conducted at the 4th International Conference on Conservative Management of Spinal Deformities said "The question of when to obtain radiography cannot be answered on the basis of scientific data".[26] There are currently no NICE or SIGN guidelines on appropriate treatment pathways for any type of scoliosis.

The updated search identified one Italian guideline that had performed a search on rehabilitation treatment of adolescents with scoliosis or other spinal deformities.[1] The guideline said *"there is insufficient scientific evidence for recommending or not recommending preventative school screening for idiopathic scoliosis in asymptomatic adolescents".* However, this guideline made opinion based recommendations that:

- Radiographic studies should not be ordered when the Adam's forward bend test is negative
- An ATI angle 5^{o} or 5mm of back hump should be taken as the significant cut-off points for ordering a radiographic study at initial examination. This recommendation was described as the opinion of the guideline group, as general consensus was absent.

Since the publication of these guidelines, there has been an additional review of screening programmes[20] and observational studies that have described the effectiveness a screening programme with different cut-off criteria for referral for radiography.[22,23] Further explorations of appropriate cut-off for referral are required. Cobb angle is a routine method of measuring the degree of curvature of the spine.

Criterion 8 met? No.

9. If the test is for mutations the criteria used to select the subset of mutations to be covered by screening, if all possible mutations are not being tested, should be clearly set out

Not applicable.

10. 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

What treatments do the guidelines recommend?

We looked for guideline recommendations on treatments for adolescent idiopathic scoliosis in the updated search. We focussed on conservative treatments such as exercise therapies or bracing which would be expected to be indicated for people with less severe scoliosis identified

through early detection. Surgery is indicated for more severe scoliosis where conservative interventions are not appropriate. There are limited evidence-based guidelines for the treatment of adolescent idiopathic scoliosis. In the updated search we found one guideline that had produced a mixture of evidence-based and consensus based recommendations.

Italian guidelines published in 2005.[1] A review was performed (although details of the methods of this review were not provided) and a series of recommendations made. The recommendations were classified as very strong, strong, fair and poor based on the scientific evidence; recommendations were also made on consensus or opinion, these included strong scientific consensus, fair scientific consensus, commission opinion. Of the close to 120 recommendations only 9 were based on the scientific evidence the others were consensus or opinion, reflecting the limited evidence base.

The largely consensus based recommendations include exercise therapies, brace treatment and exercise during brace wearing. The guidelines did not state which exercise programmes should be used or over what range of Cobb angles exercises are recommended. Brace treatment was recommended for people with Cobb angles over 20 ± 5 degrees. It was recommended that braces should be worn for more than 18 hours a day and the wearing time gradually reduced until the end of vertebral bone growth. The only recommendation based on strong scientific evidence (at least one RCT with results comparable with other published studies) was "mobilisation exercises should be performed to improve joint freedom of the spine braced full time but not during the release phase." However, the context and the research study which provided the evidence for this recommendation was not clearly presented in the guideline.

SOSORT consensus guidelines.[11] The SOSORT guidelines for conservative management of scoliosis recommended observation, physical therapy, scoliosis intensive rehabilitation programme, brace wear and surgery. When these different treatments are indicated is dependent on age, skeletal maturity, height as a proportion of mature height, and curvature on presentation. They have used the Longstein and Carlson[2]progression estimation formula to produce cut offs for treatment based on the likelihood of progression.

The updated search also identified management guidelines for AIS that were not explicitly based on a systematic review[27] which were written by authors from the department of orthopaedic surgery at John Hopkins Hospital They say that "in general, bracing is indicated for skeletally immature patients (Risser 0-2) who present with curves of 30 to 45^{o}". The surgical procedure, isolated posterior spinal fusion, was said to be typically indicated for curves > 45^{o} in actively growing adolescents and for curves of over 50 to 60^{o} in mature adolescents. The guideline said that anterior release and fusion should accompany posterior spinal fusion in selected patients (those who have curves > 70 or 80^{o}and curves that lack correction below 40 to 50^{o} on side bending films) and that thoracoscopic anterior spinal instrumentation is indicated primarily in single thoracic curves measuring 40 to 70^{o}.

What was the evidence for the effectiveness of treatments in 2004?

The 2004 USPSTF evidence update addressed the question "Is there new evidence that scoliosis treatments lead to better health outcomes if applied at an early stage?" This update described one RCT, 3 cohort studies 1 case-series meta-analysis. No firm conclusions were reached. It was highlighted that the quality of the studies was mixed, where adjustment for confounding may have been inadequate. They also said that the primary outcome was angle progression rather than health outcomes and that none of the studies primarily involved people detected by screening.

What is the updated evidence for the effectiveness of treatments?

EXERCISES

The USPSTF evidence update described one RCT from 1994 that compared different forms of exercise and the Milwaukee brace vs. exercise and electrical stimulation in thirty children between 6 and 16 years with Moiré angles (alternative measure to Cobb angle) between 15 and 45 degrees. The USPSTF noted that this study did not provide information on how scoliosis was initially detected. The outcome of the study was change in Moiré angle to 12 weeks of treatment. All groups showed improvement from baseline but there were no between group differences. The RCT did not include an untreated control group, therefore it is not possible to determine the expected untreated curve progression over this period.

Updated evidence base

We found three systematic reviews in the updated search that had assessed the effectiveness of physical exercises for adolescent idiopathic scoliosis, Negrini et al 2003[28]; Negrini et al 2008[29]; Fusco et al 2011[30]. These are a series of systematic reviews from the same research group. Negrini et al 2008[29] updates Negrini et al 2003[28] using the same methodology. Fusco et al 2011[30] updates Negrini et al 2008[29] using the same inclusion criteria, resulting in the inclusion of one additional study from the previous review. The inclusion criteria for these reviews were:-

- Patients: diagnosis of AIS by a specialist, confirmed through X-rays; focussed on patients in growing age (up to Risser 5)
- Experimental intervention: patients treated exclusively with PEs, without any other associated intervention
- Control group: any kind of patients, either observed or treated
- Outcome measures: only Cobb degrees: results could be reported in absolute terms or as a percentage of patients improved/worsened
- Study design: any study design

The latest review in this series was Fusco et al 2011.[30] The last review searched up to October 2009. We also looked for studies published since October 2009 in the updated search that had tested the effectiveness of exercise therapy for scoliosis against a control group or that had assessed exercise plus brace treatment compared to a control. We found one retrospective controlled study in the updated search that had assessed whether exercise in the weaning period following brace treatment could avoid loss of correction.[31] A Cochrane protocol for a systematic review on exercises for adolescent idiopathic scoliosis was registered in 2009.[32]

Effectiveness of physical exercises (PEs) vs. no treatment.

The aim of physical exercises is not only to influence spine curvature but also to increase positively the neuromotor control and stability of the spine, reduce poor posture and to increase breathing function.[30] There are numerous exercise protocols, some of which have been included in Fusco et al 2011 systematic review as appendices.[30] In their review the authors divided exercise treatments for scoliosis into exercises without autocorrection and exercises with autocorrection. Autocorrection was defined as “the ability to reduce the spinal deformity through the patient’s active postural realignment”. They said that autocorrection exercises have

to be performed in three dimensions where possible- the coronal plane to correct lateral deviation, the sagittal plane to promote physiological curvatures and in the transverse plane to reduce rotation of the vertebrae which can lead to axial plane deformities.[30] Without autocorrection was defined as no specified exercises or those with movement only in one direction.

The Fusco et al 2011[30] systematic review found studies that had looked at 7 types of exercise programmes:

- **Asymmetric strengthening PEs for correction of scoliosis** (1 RCT)

  The study compared the control condition (electrical stimulation, traction and postural training) with the intervention which was all of these treatments included in the control condition plus specific asymmetric strengthening physical exercises for scoliosis once a day for six months. There were 40 Chinese patients in the control arm and 40 in the intervention arm. The average age was 15 ±4 years and curvature was Cobb angle 24 ±12 degrees. Both groups had a significant improvement in Cobb angle in the thoracic and lumbar segments from baseline ($p<0.05$). The intervention group had a greater mean improvement (15 degrees) than the control group (7 degrees). The statistical significance of this difference was not reported in the systematic review, and the original publication for this study was not accessible as it was written in Chinese.

- **Scoliosis intensive inpatient rehabilitation (SIR) Schroth method** (2 case series and 1 prospective controlled study)

  The prospective controlled study compared treatment with SIR (4 to 6 weeks of intensive inpatient SIR) to no treatment in two age groups. Follow up was 33 months. The first age-group had a mean age of 10 years and a mean curve of Cobb angle 21°. They had a 53% improvement in Cobb angle compared to 29% in controls. The older group had a mean age of 13 years and a mean curve of 29.5 degrees and the treatment group showed 70% improvement compared to 44% in the matched control group ($p<0.05$).

- **Integrated scoliosis rehabilitation (ISR)** (1 prospective controlled study)

  Includes physio-logic® exercises, 3D exercises made easy, pattern specific ADL and the Schroth method and where appropriate scoliosis-specific spinal mobilization performed by the physiotherapist on the patient. The prospective controlled study compared 18 patients who received ISR to 18 patients who received the Schroth method only (SIR) for 4 weeks. The average age was 15.3±1.1 years in the experimental group and 14.7±1.3 in the control group. The average initial curve was 34.5±7.8 degrees in the experimental group and 31.6±5.8 in the control group. Thirteen of the 18 patients in each group were also wearing a brace. Lateral deviation and surface rotation improved in the treatment group by 2.3 mm and 1.2° respectively, which was larger than the 0.3 mm and 0.8°improvement seen in the control group.

- **Outpatient Schroth programme** (2 non-controlled studies)

  One retrospective study reported on 43 patients with an initial average age of 12 years and initial curve of 19.5 degrees who had followed a 4 hour per week programme for 3 months. They found that 11.6% worsened and 44.2% improved. The second study prospectively followed 50 adolescents through a 6 week, 4 hours per day, 5 days a week programme plus 90 minutes per day home exercise programme. These adolescents

were on average 14.1 years of age and had an average Cobb angle of 26.1^{o} initially. The average Cobb angle at 1 year was 17.8^{o}. All 50 patients showed some degree of improvement.

- **Outpatient Dobomed programme** (1 non-controlled study)

  A method of physiotherapy applied to patients with idiopathic scoliosis, focuses on correction of the scoliosis by active movements of the vertebral column and an active three dimensional autocorrection. The prospective study followed 136 AIS patients aged 6 to 18 years for 12 months. The researchers reported a 31-39% decrease in the Cobb angle according to the segment of the spine considered.

- **Outpatient side shift programme** (1 prospective controlled trial comparing side shift to bracing, 2 non-controlled studies)

  Autocorrection technique using a lateral shift to of the trunk to the concavity of the curve. This technique aims to reduce lateral tilt at the inferior end vertebra and to correct the curve in the side shift position. The prospective controlled trial included 164 patients, 44 adolescents received 10 to 12 half hour sessions once a week to learn the side-shift technique and were then asked to remember to use the technique as often as possible over a 2.2 year period. One hundred and twenty adolescents wore a brace for a three year period. At the start of the study the average age was 13.6 and the average Cobb curvature was 20 to 32 degrees. There was no statistically difference between the two groups, the brace group showed on average a 0.5 decrease in Cobb angle, the side shift group showed a 2.6 degree progression.

- **Outpatient Scientific Exercises Approach to Scoliosis (SEAS)** (6 controlled studies)

  Autocorrection programme where autocorrection is performed by the “patient exclusively through the spinal deep paravertebral musculature in all three planes, without external help, thus pursuing the precise control of movement without using muscular contractions strategies that drive the spine into passive alignment”. Increasing spinal stability is one of the primary therapeutic goals of the SEAS approach because the scoliotic spine is characterised by an intrinsic instability. The outcome of the 6 controlled studies are summarised in Table 2 below (based on the information provided in the systematic review; for several of these studies we were unable to retrieve the full text to clarify information which was not described in the review).

Table 2: A summary of the studies assessing the SEAS exercise programme for AIS. Reported in Fusco et al 2011[30]

Study	Study type	Average age of participants	Initial curvature	Treatment (n)	Comparison (n)	Follow up	Outcome
Mollon and Rodot 1986 In French	Not clear, not available via PubMed	10.1 years	Not clear	SEAS Lyon method (160)	Observation (50)	End of treatment (4 years)	Treated patients 63% improved, 34% worsened and 3% unchanged. Observation only 20% improved, 75% worsened 5% unchanged
Ducongé 2002 In French	Not clear, not available via PubMed	Not clear	Not clear	SEAS Lyon (422) duration of treatment unclear	Observation (169)	Not clear	Treatment improved 58% worsened 42%. Observation improved 23% worsened 77%
Ferraro et al 1998	Not clear, not available via PubMed	Not clear	Not clear	Many methods, high compliance (greater than 30 minutes a day)	Many methods, minimal compliance (less than 10 minutes a day)	Not clear	Maximal participation "slowed down or even halted the progression of scoliosis" (difference between the groups: 9 degrees)
Negrini et al 2006	Prospective controlled trial	12.4 years	15 degrees	SEAS 1 year (48 participants in total)	Usual physiotherapy	1 year (need for braces)	Failure of treatment- need for braces 4.3% SEAS vs. 20% usual physiotherapy (p=0.07) Improvement (in curvature) 28.9% SEAS, 5% in controls.
Negrini et al 2008	Prospective controlled cohort	12.4 ± 2.2 years	15 ± 6 degrees	SEAS (35)	Usual physiotherapy (39)	12 months	Need for braces 6.1% in SEAS group vs. 25.0% in the usual treatment group. Cobb angle improved in 23.5% of the SEAS group,

Study	Study type	Average age of participants	Initial curvature	Treatment (n)	Comparison (n)	Follow up	Outcome
							worsened in 11.8%. These changes were not considered clinically significant Physiotherapy improved 11.1%, worsened 13.9%
Negrini et al 2006	Prospective controlled study	13.5 ± 2.4 years	31.1 ± 11.1 degrees	SEAS plus brace (40)	Brace (70)	5 months	SEAS plus brace 58% improved, 1.5% worsened Brace: 45.8% improved, 10.3% worsened. Clinical results were defined as variations of at least 5 Cobb degrees, or 2 Bunnell degrees (measured using a Bunnell scoliometer)

The review also assessed exercise programmes which did not involve autocorrection. They looked at asymmetric and symmetric exercises.

Two non-controlled observational studies had described asymmetric exercises. Both studies tested the MedX Rotary Torso Machine. The first study involved 12 patients with an average age of 13.1 years and 33.5 ± 12.2 degree curves. After 4 months curves improved by 19% with the mean curve reduced to 27.2 ± 14.7 degrees (one patient's curve worsened). The other study involved nine 14 year old children with 29 degree curves who were again followed for 4 months. In this study, curves were reduced by 5 degrees. One study described symmetric exercises. In this study 42 patients aged between 12 and 15 years old with 10 degree curves on average, underwent a year-long program of mobilization, strengthening, and posture control according to the Milwaukee method. There was no difference in curve progression in these patients and 57 retrospective controls.

The reviewers concluded "there remains a need for further studies, especially RCTs that may improve the level of evidence regarding physical exercises. However, the present evidence is that physical exercise is an appropriate intervention for AIS even if it is not possible to support a specific exercise regimen"

In conclusion, there are new studies since the USPSTF review that have compared exercise regimens to observation However only one of these studies was an RCT which compared asymmetric strengthening training with postural and traction training to postural and traction training and electrical stimulation alone.

BRACES

Evidence base

We found four systematic reviews that assessed brace treatment for scoliosis in the updated search and two systematic reviews that assessed conservative treatments for scoliosis (see Table 3).[1,3,4,9,33-35] It was noted that the studies that have assessed braces for scoliosis to date are generally of poor methodological quality with limited RCTs. The studies have had heterogeneous populations, eligibility criteria, type of brace treatment, outcomes measures.

The systematic reviews differed in the types of study they included in their analysis (see Table 3). The Cochrane review for primary analysis allowed clinical controlled trials or RCTs only (permitting a prospective cohort for secondary analysis). The latest review by Maruyama et al 2011[4] permitted case-control studies alongside prospective CTs and RCTs, and graded studies on an 11 point methodological quality scale including points for randomisation, blinding and detailing compliance. Dolan and Weinstein allowed "clinical evaluations" concerning observation and/or bracing. Maruyama et al 2008 did not detail selection criteria for study type. Lenssinck et al 2005[3] included clinical controlled trials or RCTs. Weiss and Goodall included prospective controlled trials, and RCTs alongside meta-analyses.

Evidence of effectiveness of brace treatment: brace vs. observation

To assess the evidence that brace treatment was effective we looked at studies identified in these reviews that had compared brace treatment to observation only. The USPSTF update had identified a prospective cohort study (Nachemson and Peterson 1995[36]). This multi-centre study had been co-ordinated by the Scoliosis Research Society. The study which started in 1985 had recruited members of the Scoliosis Research Society who were from centres that were known to

manage many patients with adolescent idiopathic scoliosis. They included only physicians who "firmly believed in the effectiveness of treatment with a brace for adolescent girls who have idiopathic scoliosis and those that firmly believed that bracing was ineffective". Using this method they included 5 centres with 130 patients that observed their patients only and 3 centres with 115 patients that used braces. The criteria for bracing were not documented for the centres that used braces. Eligibility criteria for the whole study included having a curve which comprised at least five vertebrae and averaged between 25 and 35°. The study had also included 2 centres that used electrical stimulation but this was dismissed as ineffective and discontinued. The patients were followed until skeletal maturity or until progression (up to 4 years). The results showed that braces limited progression compared to observation or electrical stimulation. The Cochrane review by Negrini et al 2010[9] reported from this study that after 3 years the success rates (defined as less than six degrees increase in the curve) were 80% for bracing (95% CI 66 to 88%); 46 % (95% CI 25% to 56%) for observation and 39% for electrical stimulation (95% CI 19% to 59%). The rates at four years were 74% (95% CI 52 % to 84%), 34 % (95% CI 16% to 49%) and 33% (95% CI 12% to 60%), respectively (log rank test p<0.0001). This study was also included in the Maruyama et al 2011,[4] Weiss and Goodall,[35] and Lennssinck et al 2005.[3]

Since the USPSTF report there have been limited studies that have compared brace treatment to observation. In 2007 a follow up study from the Nachemson and Peterson 1995 study was published. This study published by Danielsson et al 2007[37], was reported in the Cochrane review and in Maruyama et al 2011 review and followed a subset of the original participants for on average 16 years. The participants that had received brace treatment had all attended a centre that used the Boston brace. The study had found that in the follow up period none of the brace group experienced failure (> 6 degrees progression from initial curve size) whereas 40% of the 41 people in the observation group only had experienced this degree of progression. In the observation group 20% were braced during adolescence and another 10% underwent surgery. The Cochrane Review reported of this study that over the long term (16 years), some patients braced or observed progressed more than 5 degrees (range 5 degrees to 21 degrees) while this progression meant that these patients returned to pre-treatment levels (31.9° versus 33.0° at start), observed patients (excluding 11 who were braced and 6 who were fused during growth because of failure) showed an overall progression from the start of treatment of 4.4°. Both the Cochrane review and the Maruyama et al 2011 review highlighted the methodological limitations of this study, i.e. the lack of randomisation and the methods by which the centres were selected, but it is also worth noting that the treatment period of this cohort was in the late 1980's and the braces used may not reflect the range of brace treatments available today. A follow up study looking at the health related quality of life in a subset of participants in the long term follow up of this study found no differences in the SRS-22 QOL questionnaire or the short form 36 between groups were found (Danielsson et al 2010[38]). The Maruyama et al 2011[4] review had also cited 3 additional case control studies of low methodological quality that had compared quality of life between patients who had received brace treatment compared with those that had been observed. These studies gave conflicting evidence that brace treatment affects QOL compared to observation only.

The Weiss and Goodall review[35] suggested that subsequent RCTs of brace treatment vs. no treatment are unlikely and unethical given that prospective studies (Nachemson and Peterson 1995, and Danielsson et al 2007) have showed "the Boston brace is effective". There was one additional case control study of poor methodological quality comparing brace treatment to observation-only which was included in the Maruyama et al 2011 SR.[4] In this study Goldberg et

al 1993 compared the Boston brace with or without aspects of the Milwaukee brace to observation in a population of 64 girls with an average age of 13.1 years. Thirty two girls were treated with a brace. Their starting Cobb angle was 22.2 degrees and after 2.2 years of treatment their Cobb angle was 23.8 degrees; 2 girls (6%) had surgery. Thirty two girls were observed for 2.5 years, they had a starting Cobb angle of 22.6 degrees on average and progressed to 26.3 degrees , five (16%) had surgery. No statistical differences were found in this study.

In the updated search there was a conference abstract[39] with interim results from a prospective randomised study of idiopathic scoliosis of no-treatment versus treatment with the SpineCor brace. We were unable to find peer-reviewed published results from this study but non-peer-reviewed interim results from this study were published in the educational materials for the SpineCor brace system on the SRS website. The website documented that the study had recruited 65 patients with initial Cobb angles of between 15 and 30°, Risser 0, 1 or 2 high risk of progression (increase of Cobb angle of 5° or more within the last 6 months). There were 29 individuals randomised to be treated with the brace, aged 12 years of age with a Cobb angle of 22 ± 5°. The control group contained 36 individuals aged 12 years with a Cobb angle of 20 ± 5°. (http://www.srs.org/professionals/education_materials/SRS_bracing_manual/section13.pdf)

It was not clear how long these individuals had been treated and followed, prior to these interim results. However, it was reported that three adolescents had withdrawn from the study in both groups. The outcomes of six individuals in the SpineCor group who had been weaned from the brace were not reported. In the control group of thirty three, 12% had improved, 45% were stable and 42% had worsened. In the SpineCor group of twenty who were still in treatment 76% had improved, 14% were stable and 5% had worsened. The researchers had questioned the logic of continuing the controlled study given the difference in the proportions of individuals worsening. However, it is not possible to determine from these accounts of this small study whether the SpineCor brace is effective compared to no treatment. Additionally, the effectiveness of SpineCor vs. no treatment, has also been challenged by a case series of 12 patients treated with SpineCor brace compared to natural history data from a historical cohort.[40]

One systematic review identified, Dolan and Weinstein[33] assessed the use of braces to prevent subsequent surgery compared to observation. The review determined relevant outcomes to be surgery, recommended surgery or curve progression to 50°. They selected study types that were “clinical evaluations” of observation and/or bracing. The review had 18 included studies (level III retrospective cohort studies or level IV case series). The inclusion criteria for studies were that they included participants aged eight years or older with AIS, and included participants who met current indications for brace initiation in the US which were a Cobb angle between 20°and 45°, age below 15 years and Risser 0, 1, 2. Studies had followed up to at least skeletal maturity (It should be noted that progression and surgeries could occur after this endpoint and therefore estimates of surgery rates should be considered minimum estimates). The surgical indications differed between studies and these suggested indications included progression, or specifically a curve of greater or equal to 45° or greater or equal to 50°. In one study the indication was a curve of greater or equal to 40°, pain, deformity and/or thoracic lordosis. A curve of greater or equal to 40 degrees, thoracic lordosis, failed brace treatment or unacceptable deformity were indications for surgery in another study. It was noted that 40° is not in itself an indication for surgery, neither is a lesser Cobb angle an absolute bar. Some of the papers included in this review did not report their surgical indications.

The pooled surgical rates for observation vs. brace were 22% (95% CI ,16% to 29%) range 13% to 38% for observation and 23% (95% CI 20% to 24%) range 1% to 43% following brace treatment. The brace rates are pooled from 15 studies (n=1814), the observation rates are from 3 studies (two centres; n= 139). Rates of surgery differed greatly between the studies.

The review also looked at risk factors for surgery. They assessed degree of curvature at baseline, type of brace, curve type, Risser sign, and "dose of wear" (i.e. number of hours the brace was worn and duration of treatment with the braces). When the studies were categorised by the degree of curvature at baseline, the surgery rates for the observation group who had a Cobb angle of less than 30^{o} were 12% (95% CI 6% to 23%) in people with this degree of curvature that had received brace treatments, the rates were 5 % (95% CI , 3% to 9%). In people with more than 30^{o} curvature at treatment initiation, surgery rates in the observation groups were 14% (95% CI 6% to 29%), and in the bracing groups were 34% (95% CI 27% to 28%).

No clear pattern was evident in four studies that had grouped individuals with thoracic, thoracolumbar, lumbar and double major curvatures. Individuals who began bracing at Risser 0 or 1 had a higher rate of subsequent surgery (25%; 95% CI 21% to 30%) than those braced at Risser 2 (9%; 95% CI 4% to 18%).

Dose of wear was confounded by brace type in several studies, and this data was excluded in pooled estimates for dose. The braces included the Boston, Boston/Charleston or TLSO, Charleston or Providence, TLSO or Rosenberger, Wilmington. The pooled estimates of surgery with full time wear were 22.91%, 95% CI 21% to 26% which was only slightly lower than part time wear 26%; 95% CI 19% to 34%.

The researchers said that their evidence was grade D and concluded according to the Oxford Centre for Evidence-based Medicine Levels of Evidence that it had "troublingly inconsistent or inconclusive studies of any level". A point that was raised in Weiss and Goodall[35] is that the outcome of surgery is a parameter with a known great range of variability therefore "the only justified conclusion to be drawn is that rate of surgery cannot be used to generate any valid evidence".

We additionally looked for studies from the updated search that had compared the clinical effectiveness of bracing relative to observation that had been published since the end of the search date for the most recent systematic review in this area, Maruyama et al 2011, that had searched up to November 2009. We found no subsequent controlled trials that had made this comparison. We did not assess studies that looked at compliance to brace treatment, studies that had compared the effectiveness of different brace types, bracing treatment regimens or rates of compliance to each other

In conclusion, there is no new evidence from good quality studies for the effectiveness of brace treatments compared to observation since the USPSTF report in terms of preventing curve progression or improving health outcomes.

A summary of the SRs and one guideline is given in Table 3 below.

Table 3: A summary of the systematic reviews and one guideline that have assessed brace treatments for AIS

Review	Search dates	Study types included	Conclusions of reviewers
Cochrane review Negrini et al 2010[9]	July 2008	Searched for studies that had a control group (RCT, CT) Included one RCT (comparison of brace types) and one prospective cohort study included for secondary analysis- Nachemson and Peterson)	There is very low quality evidence in favour of using braces making generalisation difficult. Limitations to the review include scarcity of data and that all included studies involved only female participants
Dolan and Weinstein 2007[33]	Not stated	Clinical evaluations of bracing and observation. 18 included studies (level III retrospective cohort and level IV case series)	This review did not demonstrate an advantage to bracing over observation in terms of surgical rates
Maruyama et al 2008[34]	July 2007	Clinical trials with controls and clinical trials with historical controls and clinical case series. The review included no RCTs. The majority of the 11 studies were case series. One study had compared	"Brace treatment is effective even in the long-term for the treatment of adolescent idiopathic scoliosis"

Review	Search dates	Study types included	Conclusions of reviewers
		observation to brace treatment	
Maruyama et al 2011[4]	November 2009	20 studies including randomised controlled trials, non-randomised clinical controlled trials, or case-control series. There were two CCTs and 18 case control studies. Two CCTs and 4 case control studies had compared observation to braces.	The results from the systematic review are difficult to interpret. There are quite a number of varying parameters between studies that make it difficult to reach any firm conclusions and the quality of information is limited
Weiss and Goodall 2008*[35]	Not stated	For braces vs. observation two level II prospective controlled trials (Nachemson and Peterson 1995 and its follow up study Danielsson et al 2007)	"There is evidence of a better scientific standard supporting conservative treatment for AIS, including in-patient rehabilitation and brace treatment"
Lenssinck et al 2005*[3]	December 2003	Included 3 RCTs and 10 CCTs that had compared different braces or braces vs. combination of braces and exercises. Only the Nachemson and	"The power and methodological quality of the studies were low, and studies were clinically heterogeneous. Therefore, it was impossible to draw firm conclusions regarding the effectiveness of conservative treatments for adolescents with idiopathic scoliosis the effectiveness of bracing and exercises is promising, but not yet established.

Review	Search dates	Study types included	Conclusions of reviewers
		Peterson 1995 study had compared brace vs. observation.	
Italian Guidelines Negrini et al 2005*[1]	Not stated	One prospective cohort (Nachemson and Peterson 1995) was only comparative study of observation vs. bracing	Made recommendations (grade C) -Brace treatment is recommended in the conservative therapy of idiopathic scoliosis -Brace treatment is recommended in treating patients curves with a Cobb angle >20± 5^{o}, future growth potential, and demonstrated progression of deformity or elevated risk of worsening, unless otherwise justified in the opinion of a clinician specialised in spinal diseases.

* denotes that the systematic review/guideline assessed various conservative treatments

Quality of life comparison between brace treatment and observation

The Maruyama systematic review (2011) had appraised three studies that had assessed quality of life with inconsistent outcomes.[4]

We found one additional study that had determined the quality of life using the SRS-22 tool to survey patients who had been treated with braces and compared this to a sample of matched patients of a similar age and curve magnitude who were under observation only[41]. The 46 people in the brace group wore an underarm brace for at least 20 hours a day. There were 46 people in the observation group. The average Cobb angle of people in both groups was 28.8^{o}, the average age was 18 years in the observation group (ranging from 12 to 22 years) and was 16 years in the bracing group (ranging from 11 to 20 years). The bracing group had lower scores on the function/activity subscales and the self-image subscales as well as the total score on the SRS-22. Scores on the pain, mental health and satisfaction scores were similar between the two groups. When the participants were categorised by degree of curvature, the biggest difference between bracing and observation was found in those people with less than a 20^{o} curve where there were differences in function/activity, pain, self-image and total scores.

The aim of brace treatment is to prevent progression and decrease the risk of requiring subsequent surgery. In the updated search, one paper described a discrete choice experiment that surveyed patients who had been treated with braces and/or surgery and asked the 135 participants to choose between hypothetical brace treatment profiles with different effectiveness, visibility, discomfort and treatment duration attributes. The participants said that they would be prepared to be treated with a Boston brace if it reduced their risk of surgery by 53%. The patients required risk reductions of between 32 to 74 % to accept treatment duration of 3 years. The most important determinants of the choices were the effectiveness of the brace and the discomfort of wearing a brace.[42]

Planned randomised controlled trials to compare braces with observation.

Good quality controlled trials of brace treatment compared with observation are key to being able to determine the best management strategy for adolescents with idiopathic scoliosis, but concerns have been raised as to the acceptability of not offering a treatment to patients in these trials.[35]

This issue is being addressed. There is currently a randomised controlled trial (BrAIST) of bracing supported by the National Institutes of Health/National Institute of Arthritis and Musculoskeletal and Skin diseases. This trial has a ClinicalTrials.gov identifier NCT00448448 and will compare a TLSO brace worn for at least 18 hours a day to watchful waiting.[43]

Additionally the study design for a Dutch trial has been published which aims to compare bracing using a Boston brace with observation (Netherlands trial register ISRCTN36964733).[44]

SURGERY

Dolan and Weinstein[33] highlighted that there is no established Cobb angle cut-off where patients are indicated for surgery. Out of 18 studies they found that surgical indications differed between studies.

Weiss and Goodhall[35] highlight that surgery is not indicated solely on Cobb angle and included in their systematic review an illustrative photograph of a 14 and a half year old girl with a minimal

risk of progression and where despite having a Cobb angle of more than 40^{o}, clinical scoliosis was not visible owing to the position and pattern of the curve.

Surgical treatment guidelines[45] suggest curve classification criteria prior to deciding on intervention. These guidelines describe the Lenke classification system which is an alternative to the King Moe classification which analyses thoracic curves and only addresses coronal plane deformity.

The evidence for the effectiveness of surgery is limited. The USPSTF update described one retrospective cohort that had compared the outcomes of 142 patients who had undergone surgery and 110 patients who had brace treatment. Since this report we found one SR in our updated search that had reviewed prospective controlled studies that have considered surgery vs. observation[46]. The systematic review which was published in 2008 (with no documentation of the search dates) found no prospective controlled studies, RCTs or meta-analyses that compared surgery to observation only. The reviewer said there are level III papers to support surgery but most of these were without controls and their high drop-out rate limits the conclusions that could be drawn. The review highlighted that there are prospective controlled studies comparing the long term outcome of scoliosis surgery to bracing, but that a superior effect of surgery over bracing was not concluded by the individual study authors. The review said that based on the existing evidence base "there is no medical 'intervention for surgery'" and therefore "the indication for surgery in patients with AIS is clearly limited to cosmetic reasons". The potential harms of surgery also need to be considered alongside the general risks of surgery, we found one study from the updated search that suggested fusion for scoliosis can result in diminished spinal motion and flexibility.[47]

The cosmetic impact of scoliosis may affect psychological wellbeing in some people but other people to a lesser extent. A point that applies to all treatment strategies for adolescent idiopathic scoliosis is that different individuals are likely to be psychologically affected by their degree of deformity to varying degrees. Any screening intervention should be mindful not to promote the feeling that deformity not causing psychological or physical impairment "need" to be corrected for cosmetic reasons.

The Dolan and Weinstein review on surgery rates [33] had compared surgery rates in 3 observation cohorts compared to 15 cohorts where patients had been treated with braces. The rates of surgery differed greatly between study centres. However the study authors also noted that two of the observation cohorts (which had low surgery rates) were treated in a centre that had a policy to not advocate treatment from the start (the policy in Dublin since 1991). They suggested that this could result in less anxiety on the part of the patient and family about AIS and decrease patient demand for surgery. This hypothesis has not been tested as yet. The study authors also highlighted that future long term studies are needed to determine whether "patient-determined" rates of surgery are more appropriate than "clinically determined" rates of surgery for AIS in terms of health and function throughout adulthood.

Criterion 10 met? No. There is low level evidence for the effectiveness of conservative treatments for AIS. The evidence level is low because there are few RCTs or controlled trials that have assessed the effectiveness of braces or exercise compared to observation. This means that it is not possible to assess whether these conservative treatments prevent progression compared to no treatment. A proportion of people do not experience curve progression when management is conservative. Where comparative studies have been performed the studies that have assessed braces and exercise treatments have been small and have assessed a range of different brace types or exercise programmes, therefore it is not appropriate to pool data from

these studies to make an overall assessment of effectiveness of these conservative treatments. The USPSTF update had noted that none of the studies included in its review had assessed the effectiveness of conservative treatments for AIS included participants that had been detected by screening. Although screening may be expected to detect a proportion of people who have scoliosis that is not severe enough to be indicated for surgery but may receive conservative treatments, no studies have assessed whether screen detected cases fare better than non-screen detected cases following treatment with conservative treatments. In conclusion there have been additional studies published on exercise treatments for AIS and a systematic review written. However, only one of these studies was an RCT and studies of high methodological quality remain limited. There are no additional high quality studies that have assessed bracing or surgery since the 2004 review, however there are two RCTs comparing observation to bracing which have been registered. There remains uncertainty surrounding treatments for AIS.

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

There are no NICE or SIGN treatment guidelines for scoliosis of any aetiology. One other guideline on treatment for AIS had performed a systematic review and was identified in the updated search[1], these predominantly made consensus based recommendations. There are uncertainties surrounding the effectiveness of treatments for scoliosis which would need to be resolved before an evidence based policy for treatment could be made.

Criterion 11 met? No.

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

The uncertainties surrounding treatments and the lack of evidence-based policies on the management of scoliosis suggest that this criterion is not met.

Criterion 12 met? No.

13. There should be evidence from high quality Randomised Controlled Trials that the screening programme is effective in reducing mortality or morbidity. Where screening is aimed solely at providing information to allow the person being screened to make an "informed choice" (eg. Down's syndrome, cystic fibrosis carrier screening), there must be evidence from high quality trials that the test accurately measures risk. The information that is provided about the test and its outcome must be of value and readily understood by the individual being screened

Is there a controlled trial that has compared screening to no screening?

The systematic review that had looked at the effectiveness of school screening by Fong et al 2010[20] included retrospective cohort studies only. The outcomes that it reported were the prevalence of increasing degrees of curvature and treatment for scoliosis, referral rates and positive predictive values. We wanted to address the questions:

- Would clinically significant scoliosis (defined by Cobb angle), or scoliosis requiring treatment be detected through usual care?
- Would screening detect additional people who would benefit from early treatment over usual care?

The USPSTF had reported on a prospective Dutch cohort that assessed whether addition of an annual screening programme to the usual biennial health check-up had additional benefits. The study identified 57 out of 30,000 children who had gone through the screening programme that had AIS. Thirty four children were diagnosed with AIS outside of screening. The review said that "the results showed that adolescents who had more severe cases of idiopathic scoliosis were, for the most part, previously detected outside of this screening programme (during prior school health checks)".

Since this update we found two additional Dutch papers in the updated search that have assessed school screening for scoliosis. In the Netherlands it is estimated that 80% of children are screened for adolescent idiopathic scoliosis at least once [48]. The first was a retrospective follow up study of people who had been treated with a brace, surgery or brace followed by surgery for AIS. Of the 125 patients, 66 had been detected through screening (between ages of 11 and 14), 53 were detected otherwise and for the remaining it was either unclear or not recorded. The programme sensitivity was 66/119 (55%) The authors found that for the total group the mean Cobb angle at detection was 28^{o} (SD 12.6^{o}) for the screen detected population and 40^{o} (SD 15.7^{o}) for the otherwise detected population. The differences between Cobb angles was only different for individuals diagnosed ≥ 11 years (screen detected 32^{o} (SD 11.9^{o}), otherwise detected 44^{o} (SD 15.6^{o})). For children under 11 years the average Cobb angle was 25^{o} (SD 12.3^{o}) in the screen detected population and 29 (SD 12.5^{o}) in the otherwise detected population. In total 45% of the screen detected patients needed surgery compared with 75% of the otherwise detected patients (odds ratio 0.27; 95% CI 0.12 to 0.60)). This study was small and is subject to bias which may have overestimated the effectiveness of screening. For example scoliosis patients who have a large curvature at a young age may be at greater risk of progression requiring surgery and may have had a shorter preclinical phase during which they could be detected. Screening may have also increased the likelihood that people would have visited a specialist and been prescribed a brace when their Cobb angle was smaller. Whether wearing a brace decreases the rate of surgery for scoliosis is uncertain (discussed in Criterion 10 below). Because of the potential sources of length bias and over-treatment bias the researchers concluded that the effectiveness of this screening programme still needs to be established.

A second Dutch study conducted by the same research group in 2008[49] was a case –control which compared 108 patients who were treated surgically for idiopathic scoliosis between January 2001 and October 2004 to 216 age and gender matched controls. Similarly to the previous study they found that screen-detected patients had significantly smaller Cobb angles at diagnosis than those detected otherwise. Screen detected patients had an almost threefold greater chance of being treated with a brace before surgery (OR 3.1; 95% CI 1.3 to 7.0). They found that 32.8% of the surgically treated patients had been screened between 11 and 14 years of age, compared with 43.4% of controls and 28% of the surgery-treated scoliosis patients were diagnosed before 11 years of age prior to the screening programme. The study authors concluded that "our results showed no evidence that screening for scoliosis reduced the need for surgery. Abolishing screening seems justified, especially because the effectiveness of early treatment with bracing is still strongly debated".

What degree of curvature would present with/without screening?

A cross-sectional study published in 2006[50] looked at referrals of 100 consecutive patients to a UK scoliosis clinic in London to determine the age when the deformity was first noted, the Cobb angle at the time of diagnosis and the person who first noticed the deformity. There were 81 females and 19 males with a mean age of 13.8 years (range 10 to 16 years). A higher proportion of cases had their scoliosis noted by family and friends but these people tended to have greater curves (70% of the referrals first detected by family and friends had Cobb angles in excess of 40^{o}). The authors said that with Cobb angles of this magnitude, non-operative measures are ineffective and say that untrained eyes are only likely to detect rib hump or spinal deformity at a later stage. The results of this cross-sectional study are summarised in Table 4.

Table 4: A summary of degree of curvature at presentation at a scoliosis clinic depending on how AIS patients are initially detected. Reported in Ali Fazal and Edgar 2006[50]

	Family and friends	**GPs or incidentally by another doctor**	**School screening**	**Teachers, mainly PE teachers**
Proportion of patients	63%	26%	8%	3%
Mean Cobb angle (range)	54^{o} (10-98^{o})	43^{o} (12-58^{o})	22^{o} (10-30^{o})	25^{o} (16-37^{o})
Proportion of patients with a Cobb angle > 40 degrees at diagnosis	70%	46%	0%	0%

Appropriate referral rates with and without screening programmes

In Canada in the 1970s a school screening programme was introduced which used the Adam's Forward Bend Test, carried out by a trained nurse in back examination, to screen children aged between 10 and 14 years for scoliosis. After the test children with suspected scoliosis were referred for further clinical evaluation. However, since 1979 these school screening programmes were progressively discontinued as it was determined by the Canadian Task Force on the Periodic Health Examination that it was not cost effective.[16] A Canadian cross-sectional study published in 2007 looked at the appropriateness of referrals for scoliosis since the discontinuation of school screening programmes in Canada using a cohort referred for an initial visit at an orthopaedic outpatient clinic in Montreal, Quebec, Canada between 2003 and 2004.[16] The study compared the rates of appropriate, non-appropriate and late referrals for patients with suspected AIS, to historical cohorts from the same region when school screening was in practice (summarised in Table 5).

The diagnosis of AIS was defined as at least one clinically significant curve of a Cobb angle of more than 10^{o} on a posteroanterior radiograph and presenting either a documented aetiology qualified as an idiopathic or no mention of other possible aetiologies for their scoliosis. Inappropriate referrals were patients who had a Cobb angle of less than or equal to 10^{o}. Late referrals were either immature patients (Risser sign of 0-3) with a Cobb angle > 30^{o} or patients

presenting with a Cobb angle of >40^{o} regardless of skeletal maturity. These criteria meant that late referrals met indications for immediate treatment with a brace or surgery. Of the 489 patients referred to the clinic 39.3% were appropriate referrals, 42.1% were inappropriate referrals, 18.6% were late referrals. Of the 283 patients with a confirmed AIS diagnosis, 44% had curvature of 11 to 20^{o}, 23% had curvature of 21 to 30^{o}, 18% had curvature of 31 to 40 degrees, 9% had curvature of 41 to 50 degrees and 7% had curvature of over 50 degrees. Close to sixty per cent of the patients with confirmed AIS were observed, 24% were treated with braces, 7% had surgery, 3% received other treatments and 6% were discharged. A further 10 out of the 283 cases had surgery recommended in the next 3 months following the initial visit. The authors made a general comparison with referral rates reported on two cohorts that had been screened in the schools screening programme between 1975 and 1978. They found that in the screened population there were numerically higher proportions of inappropriate referrals but a numerically lower number of later referrals than the cohort that was referred post screening discontinuation.

Table 5: Comparison of the referral patterns observed during and after the discontinuation of a school screening programme for scoliosis in Quebec, Canada. Reported in Beauséjour et al 2007[16]

Study	Patients Screened	Patients referred (% of patients screened)	In-appropriate referrals (% of patients referred)	Appropriate referrals (% of patients referred)	Late referral (% of patients referred)	Late referral; Cobb angle over 30^{o}
Morais et al 1985 Quebec (Canada), academic year 1977 to 1978	29195	2868 (9.8%)	1774 (61.8%)	1066 (37.2%)	28 (1.0%)	28 (1.0%)
Robitaille et al 1984, Montreal (Canada) academic years: 1975 to 1976 and 1976 to 1977	6873	713 (10.4%)	570 (79.9%)	137 (19.2%)	6 (0.8%)	6 (0.8%)
Beauséjour et al 2007 Montreal (Canada), clinical year 2003 to 2004[16]	No screening	489	206 (42.1%)	192 (39.3%)	91 (18.6%)	96 (19.6%)

Another consideration particularly of school screening programmes is that not all people who have a screening test that indicates that they require follow up, will receive this. A retrospective review of a scoliosis screening program from the District of Columbia highlighted that for adolescents who received a positive screening test, there was a low rate of reporting for care (around 47%).[51]

Is there a consensus on screening and what level of evidence supports current programmes?

The SOSORT consensus paper[26] (described in section 6 of this document) surveyed a sample of 35 individuals to assess whether consensus could be reached on the appropriateness of school screening for AIS. There was a consensus (89% agreement) in the individuals surveyed that school screening is useful for clinical purposes. The question was phrased "*Do you believe that school screening is useful for clinical purposes, i.e. does it affect the age at which scoliosis is treated?"* There was also consensus to the question *"do you believe that school screening is a valuable undertaking, even though the aetiology of idiopathic scoliosis is not yet clear and an aetiologically-based treatment has not yet been established?"* With 91% of respondents saying yes. Seventy four per cent of respondents said that they believed that school screening was useful for academic purposes i.e. whether we can learn about scoliosis from school screening.

An information statement was published in 2008[15] which was not based on a systematic review but was endorsed by the Boards of Directors of the American Academy of Orthopaedic surgeons, the Scoliosis Research Society, the Pediatric Orthopaedic Society of North America and the American Academy of Pediatrics. The report said that for those adolescents detected with moderate scoliosis, the available data neither definitively support nor refute the efficacy of bracing. However this report did document that there is currently a randomised controlled trial of bracing supported by the National Institutes of Health/National Institute of Arthritis and Musculoskeletal and Skin diseases. This trial has a ClinicalTrials.gov identifier NCT00448448 and will compare a TLSO brace worn for at least 18 hours a day to watchful waiting.[43]

The information statement said "as the primary care providers for adolescents with idiopathic scoliosis, the AAOS, SRS, POSNA and AAP do not support any recommendation against scoliosis screening, given the available literature". However, the document also does not explicitly suggest screening should be introduced. Within this document further recommendations were made regarding the age range over which screening should take place if undertaken. They said "females achieve adolescence about two years before males and are afflicted with a magnitude of scoliosis, requiring treatment three to four times more frequently than males. As a result if scoliosis screening is undertaken the AAOS, SRS, POSNA and AAP agree that females should be screened twice at age 10 to 12 and boys once at age 13 or 14."[15]

Criterion 13 met? No. There have been no RCTs which have assessed the effectiveness of a screening programme (i.e. would screening would lead to better health outcomes than no screening, without increasing the number of people who received unnecessary treatment?). Screening programmes do exist in some countries but in other countries school screening programmes have been discontinued. The arguments that we found in the updated search literature supporting screening programmes for scoliosis were based on consensus expert opinion rather than RCT research evidence.

14. 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 updated search did not identify any relevant evidence assessing acceptability of screening to health professionals and the public.

Criterion 14 met? Not clear.

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

We were not able to find evidence that supported the effectiveness of a particular screening programme (i.e. a RCT or CT that had compared the outcomes of people who had been screened compared to people who had not been screened). It is not possible to fully determine the balance of benefit and harms without this.

There are several areas where uncertainty exists which may influence the balance of benefits and harms.

The test: The positive predictive value of the screening tests are low, this may lead to the risk that people receive unnecessary referrals. Evidence based cut-offs for treatment are not established there remains uncertainty surrounding the prediction of who is likely to have curve progression and would be indicated for treatment. This may be associated with a risk of overtreatment.

The treatment: There is uncertainty surrounding the effectiveness of treatments for AIS. This may be associated with a risk of inappropriate treatment.

Criterion 15 met? Not clear. No RCTs or controlled trials assessing the balance of benefits and harms of screening were identified.

16. 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 criteria should have regard to evidence from cost benefit and/or cost effectiveness analyses and have regard to the effective use of available resource

We found two studies that had investigated costs of school screening programmes, one of a Turkish programme[52] and one of a Hong Kong programme.[53] The Hong Kong school programme screened 115,190 screened students who were in Grade 5 in 1995/96 or 1996/97. The study calculated the average costs spent on screening, diagnosing, following to age 19 years and treating. The costs were expressed in 2005 US dollars. They found that the average cost of screening, diagnosis and medical care per student screened was USD $54.64. The cost of finding one student with a curvature of ≥20^{o} was USD $4475.67 and the cost for finding one treated student was USD $20,768.29. The screening only costs (without diagnostic costs or medical care cost were USD $17.94 per student. The screening tests included a first tier forward bend test in the Student Health Service Centre, followed by more frequent second tier screening, conducted in a Special Assessment Centre which performed the forward bend test including measurement

of the angle of trunk rotation, Moiré topography and assessment of clinical signs. The Turkish study[52] assessed prevalence of scoliosis identified through a forward bend test by one orthopaedic surgeon, one resident and one nurse in a sample of 4259 school children aged 10 to 14 years in Istanbul. The study assessed screening costs, including diagnostic costs, salaries, transportation, communications and filing costs. They found that screening was associated with a cost of USD $0.47 per child; the cost per case of confirmed scoliosis was estimated at USD $236.81 and cost per case of scoliosis brought to immediate treatment was USD$1302.5, this excluded one child who had previously undergone surgery for their scoliosis. In total, two out of the eleven children who were found to have radiographic evidence of scoliosis were started with brace therapy. No UK cost studies were identified. No cost effectiveness studies from the UK or internationally were identified.

Criterion 16 met? Not clear. No UK cost studies were identified.

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

Not assessed.

18. There should be a plan for managing and monitoring the screening programme and an agreed set of quality assurance standards

Not assessed.

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

Not assessed.

20. 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

Not assessed.

21. 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

Not assessed.

22. If screening is for a mutation the programme should be acceptable to people identified as carriers and to other family members

Not applicable.

Conclusions

Adolescent idiopathic scoliosis is a condition that can impact on quality of life. In some people scoliosis can progress from a minor to major curve over time whereas most people who present with minor curvature will not show further progression to a severe curve. A potential benefit for screening is that it might identify people with minor curves who have not noticed their curvature and could be offered treatment to prevent progression. There is evidence to suggest that screening programmes identify a higher proportion of people with minor curves than would present outside of screening, where people on average present with more major curvature. There is no high level evidence to suggest that treatment of minor curvatures will prevent progression.

The screening tests for scoliosis are simple and safe in terms of administration but poorly predictive of scoliosis requiring intervention. As referral after a positive screening test will lead to radiography, the poor positive predictive value of these screening tests will mean that a proportion of people will have follow up including X-rays they would not otherwise have had. There have been studies which have assessed different combinations of screening tests but the highest positive predictive value for clinically significant scoliosis reported from the range of screening tests offered remains under 50%.

There is low quality evidence for the effectiveness of treatments for scoliosis. There have been new studies describing exercise and brace treatments, and systematic reviews of these studies published since the USPSTF document. These studies have largely been non-comparative and of poor quality. There have been few randomised controlled trials or controlled trials. There have been no RCTs which have compared brace treatment or surgery vs. observation. There are also uncertainties as to when conservative treatments are indicated and for whom. Risk factors for likelihood of progression have been investigated, but validation studies of these risk assessment models are required. We identified no agreed evidence based recommendations on when treatment is indicated. These uncertainties mean that there is a risk that people with AIS could receive unnecessary and ineffective treatments.

There have been no RCTs of screening programmes for adolescent idiopathic scoliosis and it is unclear whether screening would result in favourable health outcomes.

Implications for policy

Owing to the remaining uncertainties surrounding the test and treatment the updated evidence does not suggest that changing the current policy would be appropriate.

Implications for research

Prior to testing the effectiveness of a whole screening programme, further research is required assessing more refined screening strategies to determine whether it is feasible to improve the diagnostic accuracy of screening tests for AIS. Further information is additionally required on the natural history and risk of curve progression for people with minor curves is required. A suitable and validated risk prediction model would inform when treatments are indicated. There are two RCTs that have been registered to compare brace treatments with observation and results of these should be considered when the trial is complete. Further comparative studies, for example RCTs, are needed to determine the effectiveness of exercise programmes relative to observation and to evaluate the outcomes of surgical management.

Methodology

Search strategy

BACKGROUND: The current policy recommendation not to screen is based on the 2004 review and recommendations of the US Preventative Services Task Force.

US Preventive Services Task Force. 2004. Screening for idiopathic scoliosis in adolescents: brief evidence update.
http://www.uspreventiveservicestaskforce.org/3rduspstf/scoliosis/scolioup.htm

[accessed 17 February 2011]

The search for this review included articled published 1994-2002. Therefore the searches for this update included articles published from 2002 onwards. The initial update search was run in February 2011 and these searches were re-run on the same databases on August 16th 2011.

SOURCES SEARCHED: Medline (OvidSP), Embase, PsychINFO, Cinahl, Web of Science and the Cochrane library.

DATES OF SEARCH: January 2002 to February 2011 (followed by updated search on August 16th 2011)

SEARCH STRATEGY:

(N.B. numbers in brackets are from first search)

1. Scoliosis/ (11803)

2. scoliosis.tw. (11106)

3. 1 or 2 (14589)

4. exp child/ (1334508)

5. adolescent/ (1358304)

6. (child$ or adolescen$).tw. (875501)

7. 4 or 5 or 6 (2270199)

8. 3 and 7 (9509)

9. Mass screening/ (68063)

10. (test or tests or testing).tw. (1176977)

11. detect$3.tw. (1187366)

12. screen$3.tw. (343820)

13. Predictive value of tests/ (105120)

14. “Sensitivity and specificity”/ (225929)

15. (sensitiv$ or specific$).tw. (2233480)

16. (false adj (positive$ or negative$)).tw. (45166)

17. forward bend$ test.tw. (49)

18. Cobb angle.tw. (1172)

19. angle of trunk rotation.tw. (26)
20. Moire Topography/ (148)
21. moire topography.tw. (30)
22. exp Questionnaires/ (223843)
23. 9 or 10 or 11 or 12 (2417729)
24. 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 (2613025)
25. 23 and 24 (725407)
26. Spinal fusion/ (13394)
27. Braces/ (3859)
28. Casts, Surgical/ (6972)
29. spinal fusion.tw. (2950)
30. brace$.tw. (56922)
31. cast$.tw. (56922)
32. "Quality of life"/ (96185)
33. Treatment outcome/ (445582)
34. Disease progression/ (71933)
35. 26 or 27 or 28 or 29 or 30 or 31 (80269)
36. 32 or 33 or 34 (578969)
37. 35 and 36 (6886)
38. 25 or 37 (732085)
39. 8 and 38 (1242)
40. limit 39 to yr="2002-Current" (840)

Similar searches were also carried out in Embase, PsychINFO, Cinahl, Web of Science and the Cochrane Library.

Database	4th February 2011	August 16th 2011
Medline	840	57
Embase	867	54
Cochrane Library	164	1
Web of Science	270	8
PsycINFO	112	30
Cinahl	526	26
Total	2779	176

The above search strategies (4th Feb search) retrieved 2779 references in total. After duplicate references were removed a total of 1614 potentially relevant references were left. The title and abstracts were scanned for relevance to screening for idiopathic scoliosis in adolescents (including children under 10 years) focussing on the NSC criteria.

423 references were deemed to be relevant and are classified into the categories below according to the NSC criteria. There will inevitably be some overlap between categories.

A simple search was also carried out for ongoing trials in the metaRegister of Controlled Trials: http://www.controlled.trials.com/mrct/. Of the 142 retrieved, 1 met the criteria for inclusion and is included in the results below.

Following the updated search (16th August 2011) 176 results were retrieved. After removal of duplicates 107 references remained. These results were then sifted for relevance to the UK NSC criteria for a screening programme. 46 references were deemed to be relevant.

Systematic reviews and meta-analysis • Treatment (1) • Brace treatment (3 +1) • Surgery (5) • Physical exercise (3 +1) • The screening programme (2) • Other (+1)	**17**
Guidelines and recommendations	**6**
General, non-systematic reviews	**8**
The condition • **Prevalence/incidence (7 +1)** • **Patient characteristics (13+5)** • **Curve progression (8)** • **Progression prediction (6)** • **Quality of life (48+5)** - In non-operative treatment (11) - After surgery (20) - Comparison between treatments (6) - Comparison by age group (1) - Comparison to those with scoliosis (1) - Body image and self-esteem (4) - Patients experiences (2) - Reviews (3)	**94**
The test	**12**
The treatment • **Reviews (12)** • **Brace treatment (68 +9)**	**312**

- Current trial (1) - Reviews (12) - RCT trial design (1) - Brace management (7) - Compliance/ adherence (10) - Brace treatment with exercise (4) - Comparison of braces (5) - Night-time bracing (3) - Cheneau braces (6) - SpineCor (5) - Boston brace (3) - SPoRT concept (3) - Thermosetting plastic brace (2) - triaC brace (2) - Milwaukee brace (1) - CMCR brace (1) - Rosenberger brace (1) - Wilmington orthosis (1) - Charleston bending brace (1) • **Exercise (14 +1)** • **Non-operative treatment outcomes (17)** • **Surgical treatment (113 +12)** - Reviews (5) - Surgery after conservative treatment (6) - Anterior surgery (30) - Posterior surgery (10) - Combination of surgical techniques (7) - Comparison of surgical techniques (28) - Complications (13) - Reoperations (5) - Costs (1 +1) - Miscellaneous (8) • **Surgical outcomes (61+4)** - Radiographic outcomes (5) - Radiographic/clinical/functional outcomes (15)	

- Pulmonary function (14) - Long-term follow-up (9) - Back pain (4) - Spontaneous correction (3) - Factors affecting outcome (9) - Male-female differences (2) - Miscellaneous (8)	
The screening programme • **Screening in schools (12+1)** • **Screening in clinics (3)** • **Costs (2)** • **Reviews (2)**	**20**
Total	**470**

Additional relevant publications that were identified during the preparation of the update report were also included

Quality

Due to the large number of references, a first pass appraisal at abstract level was followed by retrieval of selected full text papers. Guidelines, systematic reviews of the evidence and studies addressing key areas of uncertainties in the previous reports were prioritised. For the assessment of treatment, randomised controlled trials, controlled trials and systematic reviews were prioritised, with observational evidence only described if considered relevant for key areas. Foreign language papers were excluded.

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