School Scoliosis Screening Programme-A Systematic Review

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Summary

A systematic review on the effectiveness and cost-effectiveness of school scoliosis screening programme was carried out. A total of 248 relevant titles were identified, 117 abstracts were screened and 28 articles were included in the results. There was fair level of evidence to suggest that school scoliosis screening programme is safe, contributed to early detection and reduction of surgery. There was also evidence to suggest that school-based scoliosis screening programme is cost-effective. Based on the above review, screening for scoliosis among school children is recommended only for high risk group such as girls at twelve years of age.

KEY WORDS:
Systematic review, scoliosis screening

Introduction

The Scoliosis Research Society (SRS) has defined scoliosis as a lateral curvature of the spine greater than 10 degrees (10°) as measured using the Cobb method on a standing radiograph. Idiopathic scoliosis is the most common form of lateral deviation of the spine with no clear underlying cause. The adolescent form accounts for the majority of cases of idiopathic scoliosis. The prevalence of adolescent idiopathic scoliosis varies according to the Cobb angle, between 0.1% in curvature greater than 40° Cobb angle to 2% to 3% in curvature greater than 10° Cobb angle.[1] Severe scoliosis may have a significant physical and psychosocial impact such as decreased pulmonary capacity, back pain and lower marriage rate.[2]

One of the most popular methods for prevention of these complications is early detection through screening of school children. Screening is the presumptive identification of unrecognized disease or defect by the application of tests, examinations, or other procedures that can be applied rapidly. Beginning 1984, the American Academy of Orthopaedic Surgeons (AAOS) and the SRS endorsed the concept of screening school children for early detection of scoliosis which may have gone unnoticed. This endorsement was based on the assumption that early detection in those children at risk for worsening would lead to institution of non-operative treatments that could have a positive impact on the long-term natural history of this disorder. Without treatment many curvatures could be expected to worsen over the long-term, with some eventually needing surgical intervention. In addition, more significant scoliosis in children who may present no symptoms, could be detected by clinical screening at the time when surgical treatment for their deformity could be performed most effectively.[3]

Many devices and techniques have been used for screening of scoliosis among school children. This includes Adams forward-bending test and quantitative evaluation of deformity such as measurement of rib hump height (RHH) using humpometer, Moiré topography, and scoliometer. However, none of these techniques are diagnostic. Radiographs are required to establish the diagnosis, aetiology and severity of spinal deformity.[4] Routine clinical screening for scoliosis continues to be controversial with less than half of the United States (U.S.) currently legislating school screening. In Japan, school screening programme for scoliosis is mandatory by law. On the contrary the British Orthopaedic Association and British Scoliosis Society conclude that it should not be a national policy to routinely screen children for scoliosis throughout the United Kingdom.[5]

At present school scoliosis screening programme is not included in the Malaysian School Health Service. As a result, cases of scoliosis are often detected late, when patients become symptomatic, and require corrective surgical procedures. A study conducted in Kuala Lumpur Hospital by Chuah *et al*. among 152 patients demonstrated that the median rate of curve progression of untreated idiopathic scoliosis curves was 7.03° per year.[6] The mean age at presentation for idiopathic scoliosis was 15.5 years (3.7 to 29.9 years) and the mean curve size at presentation was 41.6° (5° to 110°). The mean age of surgery for idiopathic scoliosis was 15.69 years (7.25 to 43.92 years), the mean pre-operative curve was 66.42° (37° to 130°) and the mean post-operative curve was 36.82° (15° to 79°).[6] A cross sectional study on screening for scoliosis among 2,630 school children aged 11 to 15 years in Kuala Terengganu was conducted between May and July 2004 by Azlin A.[7] The prevalence of scoliosis was 1.44% and increased with age; 1.36% in 11 years old and 4.14% in the 15 years old and the ratio of girls to boys was 3:2.[7]

This systematic review was conducted following a request from School Health Unit, Family Health Section, Family Health Development Division, under the Ministry of Health in view of the possibility of introducing school scoliosis screening programme as part of Malaysian School Health Service. The aim was to determine the effectiveness and cost-effectiveness of scoliosis screening among school children and to provide recommendations based on evidence to policy makers.

Materials and Methods

Electronic databases such as MEDLINE, PubMed, EBM Reviews – Cochrane Database of Systematic Reviews, EBM Reviews - Cochrane Central Register of Controlled Trials, EBM Reviews - HTA Databases, EBM Reviews - NHS Economic Evaluation Database, EBM Full Text – Cochrane DSR, ACP Journal Club and DARE were searched for published literature pertaining to school scoliosis screening programmes. Additional articles were identified by reviewing the bibliographies of retrieved articles and hand-searching of journals. Further information was sought from unpublished reports. There was no limit to the search. The following search terms were used either singly or in combination: school scoliosis screening, scoliosis screen*, school health, screening program*, scoliosis, and scoliometer.

Selection of studies

For this systematic review, we included all studies that met the following conditions: the study design had to be cross-sectional, cohort, case control, randomised controlled trial (RCT) or systematic review. Scoliosis screening should be conducted among school children using Adams forward-bending test and / or measurement of RHH using humpometer, measurement of angle of trunk rotation (ATR) using scoliometer, and Moiré topography. Data were sought for the following primary outcome measures: detection rate, frequency of idiopathic scoliosis surgery, progression of idiopathic scoliosis, number needed to screen (NNTS) to identify one child with scoliosis, costs of performing screening programme, diagnostic accuracy of different screening tests used in the screening programme and safety of radiography during follow-up. Studies were excluded if it was an adult onset scoliosis or initial scoliosis screening performed not in school but other setting such as hospitals. The titles and abstracts of all studies were assessed for the above eligibility criteria. If it was absolutely clear from the title and / or abstract that the study was not relevant, it was excluded. If it was unclear from the available abstract and / or the title the full text article was retrieved. Two reviewers assessed the content of the full text articles. Disagreements were resolved by discussion.

Quality assessment

The methodological quality of all the relevant full text articles retrieved was assessed using Critical Appraisal Skills Programme (CASP) depending on the type of study design.[8] Quality assessment was conducted by two reviewers. Disagreements were resolved by discussion. All full text articles related to effectiveness were graded based on guidelines from U.S./ Canadian Preventive Services Task Force.[9] All full text articles related to diagnostic studies were graded according to Hierarchy of Evidence for Test Accuracy Studies, CRD Report Number 4 (2nd Edition), March 2001.[10]

Data extraction strategy

Data was extracted from included studies by a reviewer using a pre-designed data extraction form (evidence table) and checked by another reviewer. Details on methods, study population characteristics, intervention and comparator, outcomes measures for effectiveness, safety, cost / cost-effectiveness and diagnostic accuracy of tests used in the scoliosis screening were extracted. The extracted data were presented and discussed with the expert committee before deciding on the eligibility of articles to be included in this report.

Results

A total of 248 relevant titles were identified and 117 abstracts were screened. After reading and appraising the full text articles, twenty-eight articles were included in the results. Eight full text articles were excluded based on inclusion and exclusion criteria and quality of the studies. The breakdowns of included studies were: 16 articles for effectiveness, six articles for economic evaluation (3 overlap with effectiveness), 7 articles on diagnostic accuracy of the screening tests and 2 articles on radiologic examination. The articles comprised one case control study, one before-and-after study, 7 cross-sectional diagnostic studies and 19 cross-sectional studies. The search did not yield any health technology assessment reports, systematic reviews or RCT related to the effectiveness of school scoliosis screening.

Effectiveness Detection of scoliosis among school children

Table I summarises the prevalence and NNTS to identify a child with scoliosis and who subsequently needed treatment in nine studies.[11-19] The table indicated that generally, the overall prevalence of scoliosis among school children aged six to nineteen years with Cobb angle of 10° or more ranged from 1% to 2.5%.[12-18] Studies done by Wong et al., Yawn et al., Morais et al., Gore et al. and Pin et al. have demonstrated that the prevalence of scoliosis decreases with increases in Cobb angle whereby the overall prevalence of scoliosis with Cobb angle of 20° or more in these studies ranged from 0.14% to 1.00%.[11,12,14,15,18]

Several studies have shown that the prevalence of scoliosis was higher in girls compared to boys.[11,15,19] Wong et al. found that the prevalence of scoliosis was low in children aged six to seven and nine to ten years but increased rapidly to 1.37% and 2.22% for girls at 11 to 12 and 13 to 14 years of age, respectively.[11] In another study, Gurr JF. demonstrated that in children with curves greater than 21°, girls predominate by 5.4:1.0.[16] Ohtsuka et al. found that girls to boys predominance of scoliosis cases for curvatures of more than 20° was 10:1 and it was the same for primary school children and junior high school children.[20] Generally, the NNTS to identify a child with a Cobb angle of 10° or more ranged from 48 to 58 and to identify one child who subsequently needed treatment ranged from 429 to 466.[12,13,14,15,18]

Few studies have demonstrated the value of school scoliosis screening programmes in early detection of scoliosis. Bunge et al. in their case control study involving 108 consecutive patients who were treated surgically for idiopathic scoliosis (cases) and 216 control subjects demonstrated that patients detected through screening had significantly smaller Cobb angles at diagnosis, compared to otherwise-detected patients with a mean of 34° ± 16.1° versus mean of 46° ± 13.3°, p<0.01.[21] They also demonstrated that these patients were diagnosed at a significantly younger age than otherwise detected patients with a mean of 10.8 yrs ± 2.6 versus mean of 13.4 yrs ± 1.7. Patients detected through screening had an almost threefold greater chance of being treated with brace before surgery [(Odds Ratio (OR) = 3.1; 95% Confidence Interval (CI) =1.3 to 7.0)].[21] Similar findings were demonstrated by them in another cross sectional study involving 125 patients with adolescent idiopathic scoliosis who had completed treatment with a brace, surgery or with a brace followed by surgery.[22] They demonstrated that patients detected through screening had significantly smaller

Cobb angles at diagnosis, compared to otherwise-detected patients with a mean of 28° ± 12.6° versus mean of 40°± 15.7°, p<0.01. They also found that patients detected by screening were significantly younger at detection with a mean of 9.9 yrs ± 2.6 versus mean of 12.6 yrs ± 2.4, p<0.01. Patients detected through screening were also significantly younger at diagnosis than otherwise–detected patients, with a mean of 10.9 yrs ± 2.5 versus mean of 13.1 yrs ± 2.5, p<0.01.22

Consequences on surgical treatment
There was inconsistency in the results of four studies. Three studies reported a reduction in surgery with scoliosis screening. In 1993, Montgomery and Wilner reported that the introduction of scoliosis screening programme in schools decreased the relative risk of progression into surgical range by a factor of eight. They obtained an eight times greater risk of deterioration to Cobb angle of 45° or more before screening period than after screening period (OR = 7.9, 99% CI = 1.6 to 36), without modifying the indications for treatment before and after the implementation of the screening programme.23

Loenstin *et al.* studied the experience in Minnesota over eight years involving a quarter of a million school children screened yearly. They reported that the percentage of children requiring

Table I. Prevalence Rates and Number Needed to Screen (NNTS)   by Sex and Cobb angle

Authors	Population	Cobb angle	Age group	Sex	Prevalence (%)	NNTS
Wong *et al.* Singapore, 2004	152,000	≥ 10°	6 to 14 years	F	0.93	
				M	0.25	
				All	0.59	169
		≥ 20°		All	0.25	392
		≥ 30°		All	0.08	1160 *(treatment)
Yawn *et al.* Rochester, Minnesota, 1999	2,242	≥ 10°	Grade 5 to 19 years	All	1.80	55
		≥ 20°		All	1.00	140
		≥ 40°		All	0.40	* 448(treatment)
Soucacos *et al.* Greece	82,901	≥ 10°	9 to 14 years	All	1.70	58 458(treatment)
Morais *et al.* Quebec, 1985	29,195	≥ 10°	8 to 15 years	All	1.76	57
		≥ 20°		All	0.34	295 429(treatment)
Gore *et al.* Wisconsin County, 1981	8,393	≥ 10°	Grades fifth to tenth for girls and seventh and eighth for boys	F	2.00	
				M	1.70	
				All	2.00	50
		≥ 20°		F	0.50	
				M	0.10	
				All	0.40	221 466(treatment)
Gurr JF. Montreal, 1977	26,947	≥ 10°	Grades seven and eight	All	2.00	
Dickson *et al.* Oxford, 1980	1,764	≥ 10°	13 to 14 years	All	2.50	
Pin *et al.* China, 1985	8,165	≥ 10°	6 to 15 years	All	2.40	48
		≥ 20°		All	0.14	817
Smyrnis *et al.* Athens, 1979	3,494	≥ 10°	11 to 12 years	F	4.60	
				M	1.10	
				All	*	*

*Information not provided or examined during study

surgery had declined from 0.017% in 1974 to 0.004% in 1979. They also reported that the mean size of the major curve at the time of operation decreased from 60° in 1971 to 42° in 1979.24 Bunge et al. in a cross-sectional study involving 125 patients with adolescent idiopathic scoliosis who had completed treatment with a brace, surgery or with a brace followed by surgery reported that 45% of patients detected through screening needed surgery, compared to 75% of the otherwise-detected patients. The OR for surgery for patients detected through screening was 0.27 (95% CI = 0.12 to 0.60).[22]

However, one case control study conducted by Bunge et al. with the aim of testing the hypothesis that screening for scoliosis is effective in reducing the need for surgical treatment found that exposure to screening at the ages of 11, 12, 13 or 14 years did not reduce the chance of surgery significantly. The OR for exposure to screening at the ages of 11, 12, 13 or 14 years and getting surgery was 0.64 (95% CI = 0.34 to 1.19, p = 0.16).[21]

Cost/cost-effectiveness

There were no studies related to the Quality adjusted life year (QALY) gained. However, there were six studies on economic evaluation related to school scoliosis screening programme which were conducted in the U.S., Canada, Sweden, Greece and Singapore. The cost of screening per child ranged from as low as USD 0.07, to as high as USD 43.7 as shown in Table II, depending on how the cost was calculated.[13,14,24-26] Thilagaratnam S conducted a cost-effectiveness analysis of school based screening and follow-up programme in Singapore with the alternative of not having the screening programme.[27] His analysis was based on direct and indirect costs. The total cost to screen, follow-up and treat (bracing for 36 school children and surgery for 21 school children) in the screening programme was SGD 1,063,010.82. It was assumed that without screening programme, all school children who had their curves braced (36 in total) would have had surgery in addition to the 21 school children who had surgery even with

Table II. Estimated cost of finding one case of idiopathic scoliosis.

Authors	Cost per child / case	US dollar (USD)
Yawn et al. 2000, Rochester	Per child screened Per child with Cobb angle ≥ 20° Per child treated for scoliosis (cost based on service charge and school cost, does not include indirect costs)	$ 24.66 $ 3,386.25 $ 10,836.00
Morais et al. 1985,	 Per child screened Per child referred for diagnostic evaluation including x-rays Per case confirmed scoliosis Per case brought to treatment (direct costing, does not include indirect costs)	Canadian dollar (CAD) (In 1997, 1 US $ = 1.17 Quebec Canadian $) $ 2.31 $ 59.60 $ 194.27 $ 3,508.49
Montgomery et al. 1990, Malmo, Sweden	Per child screened - No specific screening - Conventional clinical screening (FBT) - Combined clinical Moire Screening (include health care costs and production lost)	US dollar (USD) $ 33.90 $ 43.70 $ 27.70
Lonstein et al. 1982, Minnesota	Per child screened:- (include only salary of nurse coordinator) Per child screened {include only salary of nurse coordinator and school staff (time cost)}	US Dollar (USD) $ 0.07 0.35 (range from $0.24 to $1.75 depending size of the school and the salary scale of the screening staff)
Soucacos et al. 1997, Greece	Per child screened (cost based only on transportation)	$ 0.30

a screening programme. The total cost without the screening programme was SGD 1,358,104.80. The total cost was the sum of the direct costs of surgery and follow-up, and the indirect costs which comprised the time costs for the parents accompanying the child for surgical admission and follow-up visits. Net cost, which is the difference between the cost of the screening programme and the 'saving' in the absence of the screening programme, was minus SGD 295,093.98. A sensitivity analysis was performed by varying the numbers who would need surgery. Even if only about 65% of the 36 patients required surgery, the net cost remained negative.[27]

Diagnostic accuracy

It is evident that the value of any screening programme depends to a large extent on the accuracy of the screening tests. Few cross-sectional diagnostic studies related to Adams forward-bending test, scoliometer, Moire topography and measurement of RHH using humpometer were retrieved. Table III below shows the sensitivity, specificity, Positive Predictive Value (PPV), Negative Predictive Value (NPV) and area under receiver operating characteristic curve (AUC) of Adams forward-bending test, scoliometer, Moire topography and measurement of RHH using humpometer.

Adams forward-bending test had a lower sensitivity but a higher specificity compared to scoliometer, Moire topography and humpometer.[28,29] Since the prevalence rate of scoliosis was low, the PPV was low for all the tests ranging from 0.004 to 0.29.[28,29,30,31] All the tests had high NPV ranging from 0.97 to 1.0.[28,29,30,31] Prujis et al. demonstrated that no significant difference was found in the ability to detect scoliosis between scoliometer, humpometer and Moire topography. The AUC was 0.56 for scoliometer, 0.59 for Moire topography and 0.58 for humpometer.[4]

Safety

There have been concerns with regards to over-referrals and the effect of radiation exposure to adolescents during follow-

Table III. Diagnostic Accuracy of screening tests used in school scoliosis screening programme

Authors	Pop.	Screening test	Diagnostic accuracy
		Adams forward-bending test	
Karachalious et al. 1999, Greece	2,700		Sensitivity-0.84 Specificity- 0.93 PPV-0.13, NPV-0.99
Goldberg et al. 1995, Ireland	8,686		Sensitivity-0.83 Specificity-0.98 PPV-0.08
Lauland et al. 1982, Denmark	1,034		PPV-0.10, NPV-0.97
		Scoliometer	
Karachalious et al. 1999, Greece	2,700	ATR > 0°	Sensitivity-0.96 Specificity-0.79 PPV-0.05, NPV-0.99
Huang et al. 1997, Taiwan	34,234	ATR ≥ 5°	PPV for scoliosis ≥10°-0.28 PPV for scoliosis ≥20°-0.04 PPV for scoliosis ≥30°- 0.008 PPV for scoliosis ≥ 30°- 0.004
Prujis et al. 1995, Netherland	3,069	ATR ≥ 5°	AUC-0.56
		Moire topography	
Karachalious et al. 1999, Greece	2,700		Sensitivity-1.00 Specificity- 0.85 PPV-0.07, NPV-1.00
Lauland et al. 1982, Denmark	1,034	Asymetry of more than one contour line	
Prujis et al. 1995, Netherland	3,069	Asymetry of two line or more	
		Humpometer	
Karachalious et al. 1999, Greece	2,700	RHH ≥ 5 mm	Sensitivity-0.93 Specificity-0.78 PPV-0.04, NPV-0.99
Prujis et al. 1995, Netherland	3,069	RHH ≥ 8mm thoracic or thoracolumbar ≥ 5mm lumbar	AUC-0.58

up. Chamberlain *et al.* demonstrated that patients undergoing radiography received effective doses that were low in comparison with other types of radiographic examination.32 In another study, Manninen *et al.* demonstrated that the use of photofluorographs resulted in a radiation dose reduction of about one-half and considerable savings in direct imaging costs.[33] Karachalios *et al.* showed that during their screening program, it would have been possible to reduce radiologic examination by 89.40% if cut-off limits for referral had been used such as asymmetry of two Moire fringes, a humpogram deformity = 10mm, and 8°of scoliometer angle.[28]

Organisational perspective
Training of scoliosis screening teams in screening methods was one of the prerequisite before the programme was implemented. Children who were found to be positive during screening needed to be referred to referral clinics for further examination and follow-up.[11,12,13,14]

Discussion
Routine clinical screening for scoliosis remains controversial with some countries and organisations advocating while some are against it. There was no other systematic review, health technology assessment report or RCT retrieved on this issue. From this review, there was fair level of evidence to suggest that the prevalence of adolescent idiopathic scoliosis was higher in girls as demonstrated by Wong *et al.* in Singapore, Gore *et al.* in Wisconsin Country, Smyrnis *et al.* in Athens, Gurr JF in Montreal and Ohtsuka *et al.* in Japan.[11,15,19,16,20] Although, there was no RCT conducted to assess the effectiveness of the school scoliosis screening programme, there was fair level of evidence to suggest that the programme contributed to earlier detection and less surgery in screen-detected patients as shown by Bunge *et al.* in the Netherlands, Montgomery F and Wilner S in Sweden and Lonstein *et al.* in Minnesota.[21,22,23,24]

The cost of school–based screening for scoliosis varied. A cost-effectiveness analysis conducted by Thilagaratnam S in Singapore suggested that school scoliosis screening programme was cost-effective.[27] A review on the diagnostic accuracy of the screening tests used suggested that Adams forward-bending test, scoliometer, humpometer and Moire Topography could be used for screening of scoliosis in school children, but may result in high false negatives and high false positives which may lead to misdiagnosis and over-referrals. Because of this, there was a suggestion to use combination of tests with a certain cut-off limits for referrals.[28] Organisational issues such as training, manpower, good referral follow-up system, treatment and funding must be addressed at all levels to ensure the effectiveness and sustainability of the screening programme. Evidence indicated that targeting the screening to high risk groups such as pre-pubertal girls was more appropriate and probably cost-effective as conducted in Denmark and Taiwan and also as recommended in the cost-effectiveness study by Thilagaratnam S.[30,31,27]

Limitations
Although the quality of the studies included in this review was satisfactory, there were some methodological limitations. Most of the effectiveness studies were cross-sectional studies, although this was the more appropriate study design for screening. The assessment of the methodological quality of these studies using CASP assessment tool was not possible due to limitations in the CASP checklist itself. For cross-sectional diagnostic studies, not all patients were subjected to the diagnostic test and the reference standard with the exception of one study by Karachalios *et al.*[28] This was to avoid inappropriate use of spine radiographs. This may have introduced verification bias. In addition, some studies did not conduct blind evaluation. Although every effort has been made to retrieve full text articles, there were six articles which the authors failed to retrieve full text.

Conclusion
In conclusion, the prevalence of scoliosis was higher in girls compared to boys and increased rapidly from eleven to fourteen years of age. There was fair level of evidence to suggest that school scoliosis screening programme was safe, able to detect scoliosis at younger age, with smaller Cobb angle and contributed to reduction of surgery. There was also evidence to suggest that school-based scoliosis screening programme was cost-effective. Tests used for screening of scoliosis among school children may lead to high false positives or false negatives. Based on the above review, screening for scoliosis among school children is recommended only for high risk group such as girls at twelve years of age (standard six). A combination of modalities of screening tests such as Adams forward-bending test and scoliometer with ATR of 7° is recommended with the aim of reducing the number of referrals sufficient for the valid cases to be treated. However, organisational issues such as training, manpower, good referral and follow-up system, treatment and funding must be addressed at all levels.

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