A population-based cohort study of 394,401 children followed for ten years exhibits sustained effectiveness of scoliosis screening

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ABSTRACT

BACKGROUND CONTEXT: The value of scoliosis screening has been recently shown in a multicentre randomised controlled trial. However, the long-term sustainability of the clinical effectiveness of scoliosis screening as a routine health service remains unknown.

PURPOSE: To assess the sustainability of the clinical effectiveness of school scoliosis screening.

STUDY DESIGN/SETTING: A large population-based cohort study with a ten-year follow-up.

PATIENT SAMPLE: Three hundred ninety-four thousand four hundred and one students who were in fifth grade during the five academic years from 1995/1996 to 1999/2000 formed five consecutive annual cohorts. The students were eligible for the Hong Kong scoliosis screening programme, with their screening history and medical records until their nineteenth birthdays being assessed.

OUTCOME MEASURES: Development of adolescent idiopathic scoliosis by the 19 years of age and the Cobb angle.

METHODS: The clinical effectiveness of scoliosis screening was assessed by referral rate for radiographic diagnosis, sensitivity, specificity, and predictive values. Funding Source: Central Policy Unit of the Government of the Hong Kong Special Administrative Region and the Research Grants Council of the Hong Kong Special Administrative

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RESULTS: A total of 306,144 (78%) students participated in scoliosis screening, which used a two-tier system. The prevalence of curves ≥ 20° was 1.8% (95% confidence interval [CI] 1.7%–1.8%), while the referral rates for radiography, the sensitivity, and the positive predictive value (PPV) for curves ≥ 20° were 4.1% (95% CI 4.0%–4.2%), 91% (95% CI 90%–92%), and 40% (95% CI 39%–41%), respectively. Across the five consecutive annual cohorts, the prevalence and sensitivity for curves ≥ 20° increased by 0.23% (95% CI 0.21%–0.25%; $p<.001$) and 0.76% (95% CI 0.43%–1.04%; $p<.001$) per year, respectively; however, the PPV was reduced by 1.71% (95% CI 1.09%–2.33%; $p<.001$) per year.

CONCLUSIONS: This report describes the first large population-based study with a long-term follow-up indicating that a scoliosis screening programme can have sustained clinical effectiveness in identifying patients with adolescent idiopathic scoliosis needing clinical observation. As the prevalence of adolescent idiopathic scoliosis increases, scoliosis screening should be continued as a routine health service in schools or by general practitioners if there is no scoliosis screening policy.

Keywords: Adolescent idiopathic scoliosis; clinical effectiveness; scoliosis screening, sustainability

Introduction

School scoliosis screening has been used to identify individuals with adolescent idiopathic scoliosis (AIS) who require clinical follow-up or conservative brace treatment, with an aim of reducing the risk of requiring invasive spinal fusion surgery [1]. However, despite its long history of more than 60 years, its use as a routine health service has been heavily debated [2]. In 2004, the United States Preventive Services Task Force (USPSTF) recommended against scoliosis screening as a routine health service, primarily because of the lack of effective screening tools and inadequate evidence supporting that bracing AIS patients reduces the risk of progression and hence the need for surgery [3]. This finding led to the abolishment of screening programmes or reduced enthusiasm towards scoliosis screening around the globe [4]. However, a recent multicentre randomised controlled trial has shown that brace treatment can effectively reduce the risk of progression of AIS at skeletal maturity [5]. This confirms the value of scoliosis screening for the early detection of AIS [6].

The clinical effectiveness of scoliosis screening has been assessed in numerous studies of different designs, which have been synthesised in a systematic review and a meta-analysis [7, 8]. The systematic review covered 28 studies published between 1977 and 2004 and concluded that there was sufficient evidence to suggest that school scoliosis screening is safe, may detect cases of AIS at early stages, and may reduce the risk of surgery [7]. In contrast, the above-mentioned meta-analysis focused on 36 retrospective cohort studies that were published from 1977 to 2005 and concluded that there was substantial heterogeneity across the studies, that the use of the forward bending test (FBT) alone is inadequate, and that there is a need for a large retrospective cohort study with sufficient follow-up [8]. To address this challenge, a large population-based retrospective cohort study was later conducted that followed 157,444 students from the age of 10 until they were 19 years old who were diagnosed with scoliosis via a two-tier screening protocol in Hong Kong [9]. The results showed that the programme was clinically effective in detecting significant curves, with a low referral rate for radiography and with a cost that was close to or lower than other scoliosis screening programmes [9-11]. This new evidence initiated a review of the best available evidence by an international task force, which concluded that there is scientific evidence to support the technical efficacy and the clinical, programme and treatment effectiveness of scoliosis screening [6]. However, no previous studies have assessed the sustainability of the clinical effectiveness of scoliosis screening protocols. Sustained effectiveness is a critical issue and a challenge for community health service initiatives [12]. A screening programme may be highly effective when first implemented, but its clinical effectiveness may decrease as its duration increases [13]. Inadequately designated fiscal resources, the reduced enthusiasm of the staff, unsystematic skills training, poorly articulated visions of aims and goals, and a lack of support from policymakers are some of the factors that may weaken the effectiveness of a community programme over time [12].

In Hong Kong, scoliosis screening has been conducted as a routine health service since 1995, thereby making it one of the regions with the longest history of routine scoliosis screening in the world. Hong Kong's screening protocol was demonstrated to be clinically effective for children who studied in the fifth grade during the first 2 academic years after the programme was started; however, no longer-term evaluation was attempted [9, 14]. Therefore, the aim of this study is to build on our previous work to assess the sustainability of the clinical effectiveness of scoliosis screening in a community setting for a longer follow-up period by follow-up of the children through their academic years until they reached an age of 19 years.

Materials and methods

This study was approved by the Institutional Review Board of the University of Hong Kong/Hospital Authority Hong Kong West Cluster (UW 09-113).

Study design

We conducted a population-based cohort study of students who were in the fifth grade during the academic years 1995/1996 to 1999/2000. Medical records, which included their screening history for scoliosis until they reached an age of 19 years, were retrieved. Ethics approval was obtained from the local authority.

School scoliosis screening programme in Hong Kong

Detailed procedures of the Hong Kong scoliosis screening programme have been described elsewhere [9-11]. Briefly, it is a two-tier programme. Students in tier 1 are screened in regional clinics using the FBT and by measuring the angle of trunk rotation (ATR) using a scoliometer. Those with an ATR $\geq 5°$ but $<15°$ are further screened by moiré topography, and students with a difference of two or more moiré lines between the left and right sides of the back or presenting with concomitant significant clinical signs of spinal deformity are referred for radiographic diagnosis. Those with an ATR $\geq 15°$ are referred directly to a specialist hospital for radiographic diagnosis without undergoing moiré topography. Patients diagnosed with AIS are managed and followed up in one of the only two public specialist hospitals that manage scoliosis in Hong Kong. Students in the fifth grade or those who have reached 10 years of age are eligible for screening. They are screened biennially or more frequently if there are indications of AIS at the initial screening, until they reach an age of 19 years. Children diagnosed with AIS are followed up at a specialist hospital.

The screening protocol was developed by the Department of Orthopaedics and Traumatology of the University of Hong Kong. It was first implemented in 1995 and is managed by the Department of Health of the Hong Kong Special Administrative Region Government as a voluntary programme under an annual national health assessment scheme for adolescents.

Data collection

Data were obtained from the Department of Health and the two specialist hospitals that manage scoliosis patients. We collected data on the demographics, school grades, and screening results of each visit for all fifth-grade students during the academic years 1995/1996 to 1999/2000 until they reached an age of 19 years. For the AIS patients who were followed up at a specialist hospital, their referral sources, radiographic measurements, body height, and treatment received (brace or surgery) at each clinical visit were also recorded by trained personnel. All data were meticulously noted and cross-checked with the original medical records, when necessary.

Statistical analysis

The fifth-grade students in Hong Kong in each academic year from 1995/1996 to

1999/2000 formed five annual cohorts, and the clinical effectiveness of the school scoliosis screening was assessed for each cohort. For each annual cohort, the referral rate, sensitivity, specificity, positive predictive value (PPV), and negative predictive value (NPV) for the different degrees of spinal deformity during adolescence were obtained [8-10]. The referral rate was defined as the percentage of screened students referred for radiography. The sensitivity and specificity were defined as the proportion of AIS patients in whom AIS was detected by screening and the proportion of non-AIS subjects who were negative for AIS according to the screening, respectively. The PPV was defined as the proportion of referred students with AIS detected before they reached the age of 19 years, while NPV was defined as the proportion of non-referred students who did not develop AIS during adolescence. Sensitivity and specificity are also known as accuracy measures, whereas PPV and NPV are called utility measures because while the latter pair can be influenced by the prevalence of AIS patients, the former pair cannot [15]. Note that students who were referred for radiography but did not visit a screening clinic or a specialist hospital for assessment were considered negative for AIS.

All estimates were accompanied by exact 95% confidence intervals (CI) based on a binomial distribution, and a 5% level of significance was used for all significance tests. The data management and analysis were performed using the Statistical Analysis System (SAS) version 9.1 (SAS, NC) [16].

Results

According to national official statistics from the Education Bureau of Hong Kong, 394,401 students studied in the fifth grade during the academic years 1995/1996 to 1999/2000. Their disposition in the school scoliosis screening process is shown in fig 1. A total of 88,257 students did not participate in the screening; of that number, scoliosis was detected in 126 (0.1%) before the screening, and they were therefore not screened. Among those who elected not to be screened, 515 (0.6%, 95% CI 0.5%–0.6%) of them were diagnosed with AIS by the time they were 19 years old. Among the 306,144 (78%) children who participated in the school scoliosis screening, 9,726 (3.2%, 95% CI 3.1%–3.2%) were referred for radiography by ATR or moiré topography, and 12,536 (4.1%, 95% CI 4.0%–4.2%) were referred when significant clinical signs, including uneven shoulder height, pelvic tilt, rib or loin hump, or scapular prominence and truncal shift, were detected.

Among the students screened, the prevalence rates of AIS for different curve magnitudes and treatment statuses at 19 years of age are summarised in table 1. The prevalence of AIS among the screened students was 3.5% (95% CI 3.5%–3.6%), which was significantly higher than that among students who chose not to be screened ($p<.001$). The incidence of AIS and its treatment were significantly more prevalent in girls than in boys (p<.001 using Fisher's exact test), with the ratio of girls to boys increasing with a larger curve size. The prevalence of curves ≥ 10° by the age of 19 years in the annual cohort increased linearly each year, from 2.3% in fifth-grade students in 1995/1996 to 4.7% in those in the 1999/2000 cohort, i.e., an average of 0.61% per year (95% CI 0.58%–0.64%; p<.001 using Binomial regression) (Fig. 2). Similarly, the prevalence of curves ≥ 20° increased from 1.3% for the fifth-grade students in 1995/96 to 2.2% for those in the 1999/2000 cohort, i.e., an average of 0.23% per year (95% CI 0.21%–0.25%; p<.001). This increase in prevalence was accompanied by an increase in the referral rate for radiography from 2.7% (95% CI 2.5%–2.8%) in the 1995/1996 cohort to 5.5% (95% CI 5.3%–5.7%) in the 1999/2000 cohort. Coincidentally, there was also an annual increase of 2% in the participation rate, which reached 81% of fifth-grade students in 1999/2000.

Table 2 presents the results for the clinical effectiveness of school scoliosis screening, and figs 3 and 4 depict the longitudinal changes over the five annual cohorts. For detecting curves ≥ 10°, the four measures of clinical effectiveness were generally stable, with a slight increase in the sensitivity of 0.76% per year (95% CI 0.43%–1.04%; p<.001), resulting in an increase from 92% in the 1995/1996 cohort to 95% in the 1999/2000 cohort. Moreover, both the specificity and NPV remained consistently above

99%. For detecting curves ≥ 20°, the sensitivity increased by 0.87% per year (95% CI 0.33%–1.40%; p=.001), from 89% in the 1995/1996 cohort to 91% in the 1999/2000 cohort, despite an annual reduction of 0.5% in specificity and a 1.71% (95% CI 1.09%–2.33%; p<.001) reduction in the PPV. Nevertheless, the specificity remained above 96% and the PPV remained above 37%. The NPV was consistently above 99%.

Discussion

To our knowledge, this is the first large population-based representative study to assess the sustainability of the clinical effectiveness of school scoliosis screening as a routine health service in the community. The prevalence of AIS in Hong Kong has been increasing since the screening programme began, but the sensitivity and predictive value of the Hong Kong screening programme for detecting AIS were sustained over five consecutive annual cohorts of students who were followed for at least 10 years.

A screening programme should preferably be highly accurate, with the sensitivity, specificity, PPV, and NPV as high as possible. However, the implementation of a screening programme will inevitably have suboptimal accuracy due to behavioural noncompliance. It has been recommended that the sensitivity and specificity of a screening programme should be 70% or more, while the PPV is often between 30% and 50% [17]. To detect curves ≥ 20°, a degree that merits at least clinical monitoring, the sensitivity and specificity of the Hong Kong scoliosis screening protocol were 91% and 98%, respectively, which are far above the recommended standard of 70%. Moreover, the corresponding NPV was almost 100% and the PPV was 40%, both of which fall within the commonly reported range. Hence, the Hong Kong scoliosis screening programme has demonstrated acceptable accuracy in detecting curves deserving of clinical monitoring. The PPV for detecting curves ≥ 20° in the Hong Kong scoliosis screening programme decreased over the annual cohorts. This is unlikely to be due to the influence of the corresponding prevalence, as the prevalence rate of curves ≥ 20° increased over the annual cohorts, which should have increased the PPV [15]. The reduction in the PPV for curves ≥ 20° may be a consequence of an increase in the referral rates for radiography. Given that the PPV for curves ≥ 10° was maintained at a high level and the sensitivity for detecting curves ≥ 10° or ≥20° has increased, the additional referral rate is unlikely to be a result of a decline in screening skills, but it may be due to increased concerns about the disorder as more AIS patients were identified.

The screening protocol adopted in Hong Kong continued to be more effective than other existing programs even several years after its implementation in 1995 [8]. Starting a school scoliosis-screening programme as an additional health service in the community requires investing enormous efforts not only in developing the screening protocol but also in setting up procedures across various units in the healthcare system, obtaining screening equipment, and training staff. The skill of the staff is perhaps the most highly variable factor that influences the clinical effectiveness of scoliosis screening programmes. The personnel involved in scoliosis screening may be meticulous and attentive when they have first learned their screening skills, but this skill may not be sustained over years of routine administration and staff rotation. Staff enthusiasm is another important factor for sustaining the clinical effectiveness of screening. The recommendation against scoliosis screening made by the United States Preventive Services Task Force reduced the enthusiasm for scoliosis screening [3]. This recommendation even led to the cessation of scoliosis screening by nursing associations [18]. Sustaining the clinical effectiveness of a screening programme is essential to ensure the long-term feasibility and value of screening. The success of the Hong Kong scoliosis-screening programme in maintaining an effective performance is most likely because the programme was highly centralised and was coordinated by the Department of Health with support and supervision from a University Department. This ensured strict adherence to the screening protocol, and screening personnel were properly trained before they could begin screening. The Department of Orthopaedics and Traumatology of The University of Hong Kong had been conducting regular courses for screening doctors to ensure their skills were maintained and to train new screeners. All screening personnel were also monitored for a month after training before they could screen students independently.

Participation in the Hong Kong screening programme has also been improving, from the previously reported 73% among the first two annual cohorts of students to 78% when the next three annual cohorts were also considered. This participation rate was higher than those reported in Singapore and Minnesota and is comparable to the highest reported participation rate in the literature of approximately 80% in the Netherlands [19-21]. Indeed, since the implementation of the Hong Kong scoliosis-screening program, there have been regular radio and television programmes to educate the public about the importance of scoliosis screening. The increase in the participation rate could be due to the public's increased awareness of scoliosis after the programme was implemented. The estimated prevalence rates of AIS with curves ≥ 10° and ≥ 20° by the age of 19 among students in Hong Kong have gradually increased over each annual cohort. There are several reasons for this increase. First, the scoliosis-screening programme in Hong Kong is voluntary, and the increased participation rate may reflect that there are more children who are concerned about spinal deformity being screened. Second, the increased screening sensitivity indicates that the chances of identifying an AIS patient through screening have increased. In other words, we may have approached the actual prevalence only in the later annual cohorts. Third, the increased urbanisation in Hong Kong may also have contributed to the increased prevalence of AIS. Indeed, there was also an increase in the annual prevalence of AIS among children screened in Korea, from 1.7% in 2000 to 6.2% in 2008 [22]. An earlier study in Japan reported that urbanised regions had a higher incidence of AIS than rural regions [23]. The average annual growth in the percentage of the total population living in urban areas in Korea between 2000 and 2010 was 1.22, whereas the equivalent figure for Hong Kong between 1995 and 2000 was even higher, at 2.14%, which was slightly above the global figure of 2.11% [24]. Although the association between urbanisation and the development of AIS is not fully understood, urbanisation may cause children to mature earlier due to improved nutritional standards [25, 26]. Because children who mature early have been shown to have a significantly longer pubescent period and increased height gain during puberty,[27] the risk of curve progression may be high.

Compared with our previous results based on the first two annual cohorts, the ratio of girls to boys has decreased from 2.7 to 2.2 for curves ≥ 10°, from 4.5 to 3.6 for curves ≥ 20°, from 8.1 to 5.5 for curves ≥ 40°, and from 8.4 to 5.3 for those receiving treatment. Although the prevalence of AIS has certainly increased among girls, the probability of AIS development among boys has been gradually increasing. This finding implies that the previous suggestion of the selective screening of girls only may not be appropriate, at least in the Hong Kong setting [28-30].

Whether students should be screened for AIS depends on not only the availability of a clinically effective screening program but also on the availability of an efficacious conservative treatment [31]. For a very long time, bracing has been the most commonly used option for the treatment of AIS, but strong evidence to support its efficacy in controlling curve progression has been reported only recently [5, 32]. A study that was started as a randomised controlled trial and later incorporated a preference cohort reported that 72% of braced patients had curves below 50° at skeletal maturity, which was significantly higher than the corresponding 48% of patients who were not braced (adjusted odds ratio = 4.1). Another clinical trial of a similar design but with both the randomised and preference cohorts planned as concurrent components has had its feasibility recently assessed and confirmed in the local setting in Hong Kong [33]. The strong evidence and continual efforts in support of the efficacy of bracing have strengthened the need for screening for idiopathic scoliosis so that bracing can be applied early to prevent curve progression and hence reduce the risk of surgery.

We have not examined the cost of scoliosis screening because it has been previously reported [11]. In Hong Kong, if at least 52% of the braced students would have required surgery had they not been braced, the cost saved from preventing the need for surgery could offset the costs of screening and follow-up. In contrast, 65% or more would be needed in Singapore to break even between the costs of surgery and the costs of screening and follow-up [34]. In general, it is difficult to compare costs associated with screening across countries because of the differences in their health-care systems and economies. Nevertheless, the costs of screening in Hong Kong appeared to be comparable with those in the U.S.

Conclusions

Sustaining the clinical effectiveness of a scoliosis screening programme requires not only an effective screening protocol but also concerted effort from administrators, clinicians, and all frontline staff involved in the screening. Regular training and monitoring of screening personnel to ensure the optimal delivery of screening procedures and publicity to ensure high participation rates in scoliosis screening play vital roles in sustaining the clinical effectiveness of the Hong Kong scoliosis screening programme. Despite the controversy surrounding scoliosis screening, this screening has provided information essential to the understanding of the epidemiology of AIS [35]. The increase in the prevalence of AIS in Hong Kong, together with the consistently clinically effective scoliosis screening protocol, supports the continuation of scoliosis screening in schools or by general practitioners in places without a scoliosis screening policy.

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FIGURE LEGENDS

Fig 1 School screening for adolescent idiopathic scoliosis (AIS)

Fig 2 Prevalence of patients with adolescent idiopathic scoliosis

Fig 3 Clinical effectiveness of school scoliosis screening for detecting curves of at least 10°

Fig 4 Clinical effectiveness of school scoliosis screening for detecting curves of at least 20°

EVIDENCE AND METHODS

Context

School scoliosis screening is safe and may detect adolescent idiopathic scoliosis. Bracing is efficacious in controlling curve progression in patients with adolescent idiopathic scoliosis.

Contribution

A carefully designed and conducted scoliosis screening programme can have sustained clinical effectiveness in identifying patients with adolescent idiopathic scoliosis who require clinical monitoring or treatment

Implications

Screening for adolescent idiopathic scoliosis should be continued as a national health service in schools or by general practitioners in places without a scoliosis screening policy

Table 1

Prevalence of Adolescent Idiopathic Scoliosis by the Age of 19 years

	Prevalence (Exact 95% CI)			
	Total	**Boys**	**Girls**	**Girls:Boys**
Curves ≥ 10°	3.5% (3.5%, 3.6%)	2.2% (2.1%, 2.2%)	4.8% (4.7%, 5.0%)	2.2
Curves ≥ 20°	1.8% (1.7%, 1.8%)	0.7% (0.7%, 0.8%)	2.8% (2.7%, 2.8%)	3.6
Curves ≥ 40°	0.2% (0.2%, 0.3%)	0.07% (0.06%, 0.09%)	0.4% (0.4%, 0.4%)	5.5
Treatment	0.4% (0.4%, 0.4%)	0.1% (0.1%, 0.1%)	0.7% (0.6%, 0.7%)	5.3

Table 2

Clinical Effectiveness of School Scoliosis Screening

	Positive predictive value (Exact 95% CI)	**Negative predictive value (Exact 95% CI)**	**Sensitivity (Exact 95% CI)**	**Specificity (Exact 95% CI)**
Curves ≥ 10°	81.0% (80.3%, 81.7%)	99.8% (99.8%, 99.8%)	93.8% (93.3%, 94.3%)	99.2% (99.2%, 99.2%)
Curves ≥ 20°	39.8% (38.9%, 40.6%)	99.8% (99.8%, 99.8%)	91.0% (90.2%, 91.7%)	97.5% (97.4%, 97.5%
Curves ≥ 40°	4.6% (4.2%, 5.0%)	99.9% (99.9%, 100%)	77.6% (74.5%, 80.6%)	96.1% (96.0%, 96.2%)
Treatment	8.4% (7.9%, 8.9%)	99.9% (99.9%, 99.9%)	83.3% (81.1%, 85.3%)	96.2% (96.2%, 96.3%)

Fig. 1. School screening for adolescent idiopathic scoliosis (AIS)

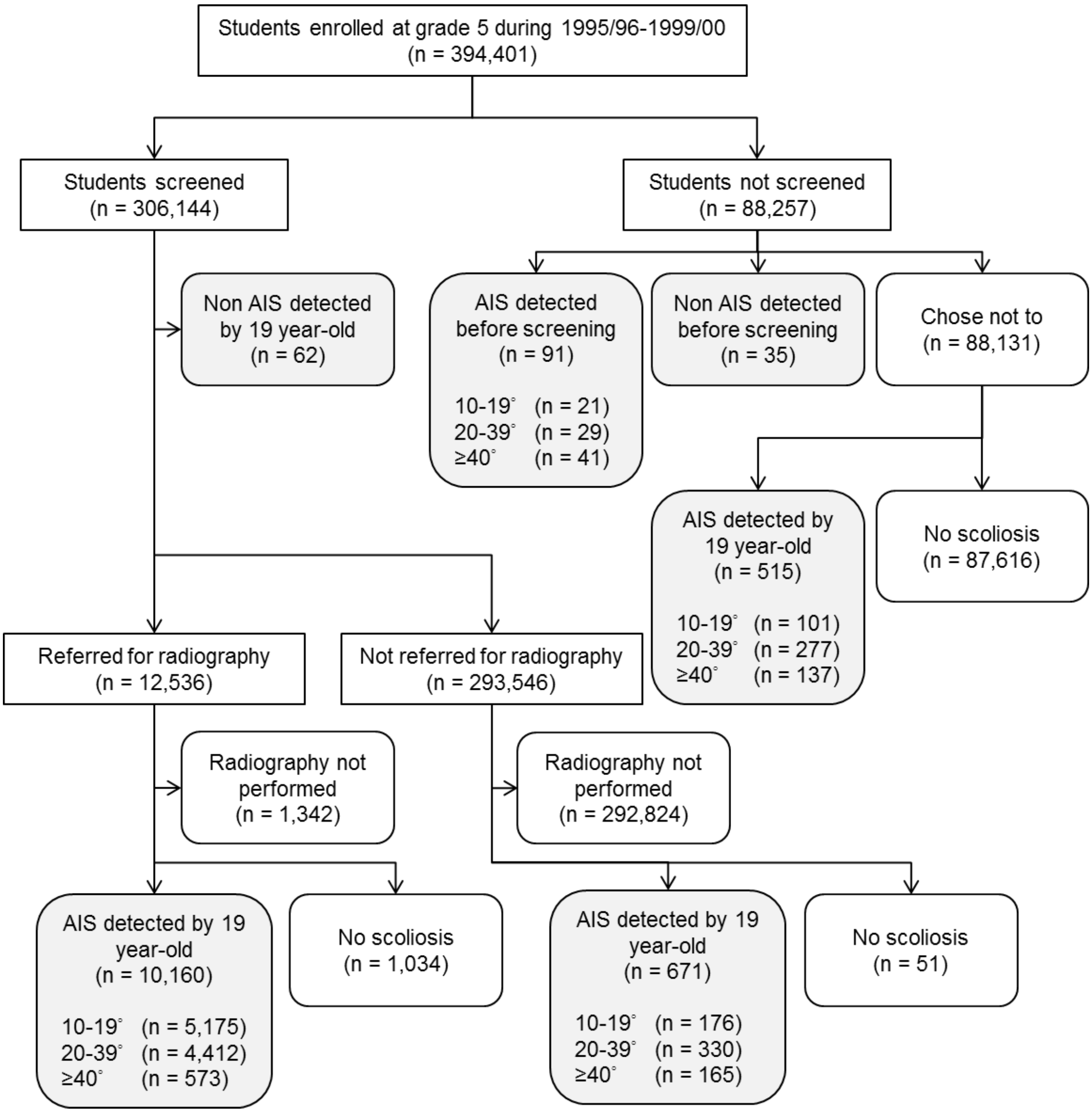
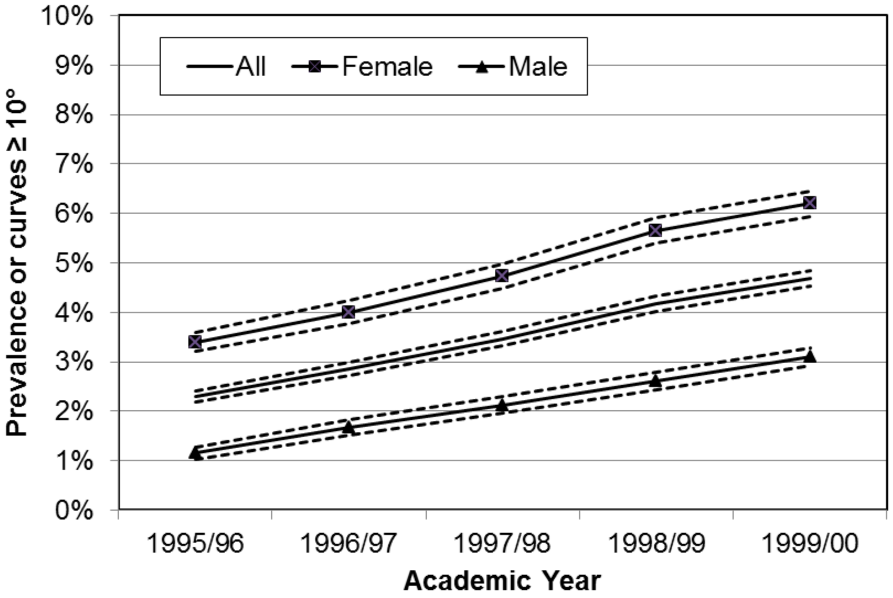


Fig. 2. Prevalence of patients with adolescent idiopathic scoliosis

(a) Cobb angle ≥ 10°



(b) Cobb angle ≥ 20°

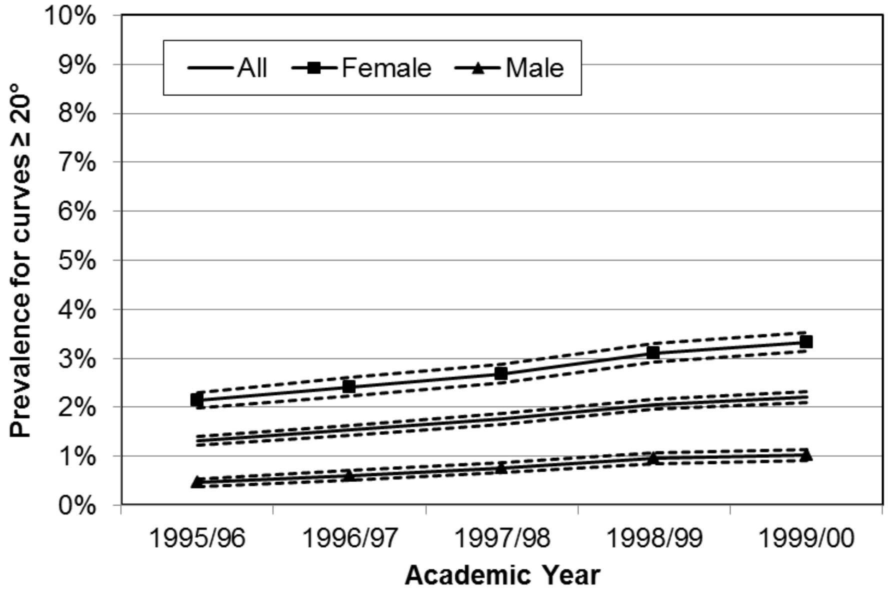


Fig. 3. Clinical effectiveness of school scoliosis screening for detecting curves of at least 10°

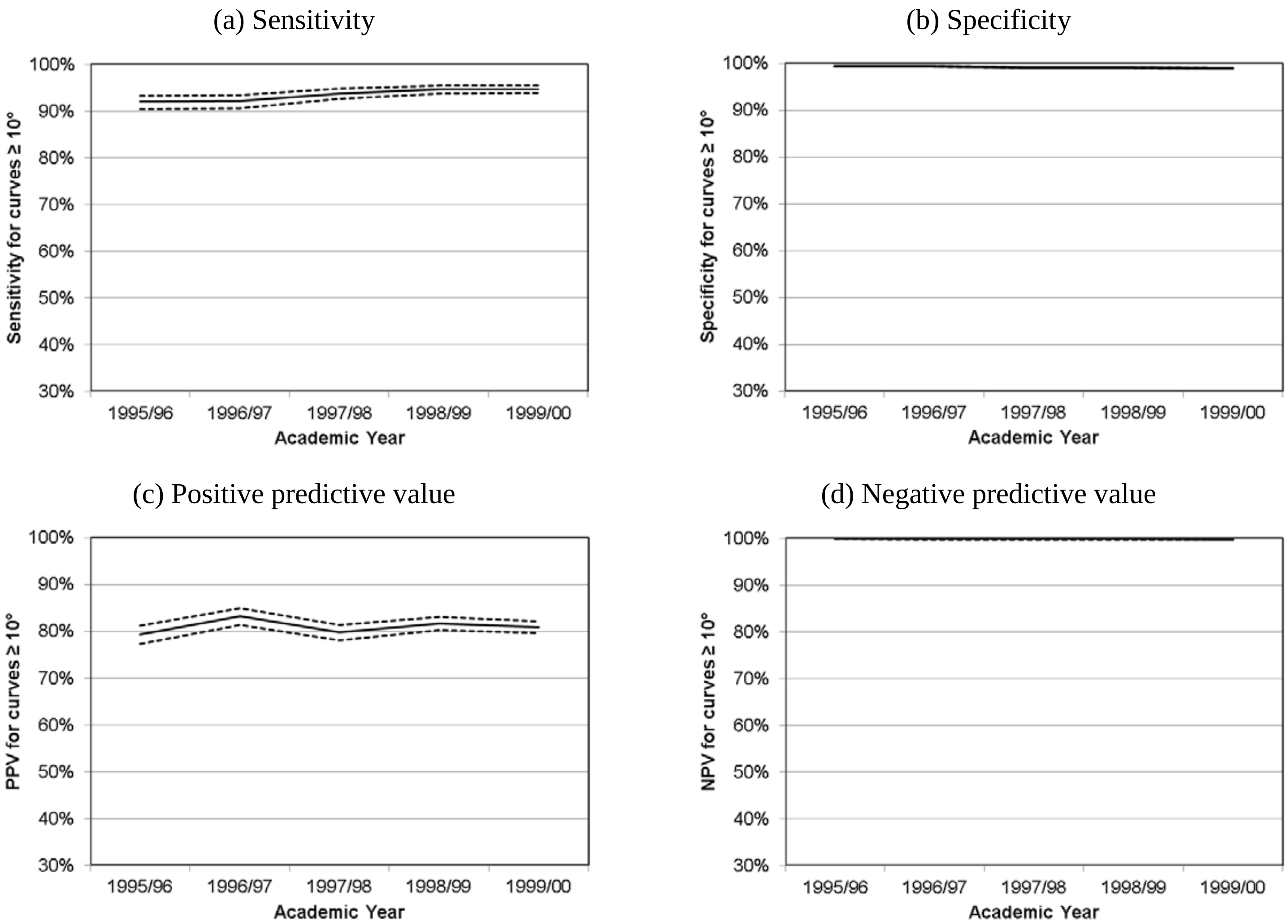


Fig. 4. Clinical effectiveness of school scoliosis screening for detecting curves of at least 20°