SCHOOL-BASED SCREENING FOR SCOLIOSIS

A Thesis

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Master of Science in Health Science

by

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ABSTRACT

Problem: Scoliosis is a spinal deformity characterized by abnormal lateral curvature that may, in some instances, progress with the child during periods of growth. In order to halt further progression of abnormal curvatures, early intervention is required. Inadequate treatment in early stages may increase morbidity and mortality in affected patients, and may require extensive invasive treatments in the future. While several governing bodies recommend the screening of children and adolescents, the United States Preventive Services Task Force (USPSTF) currently recommends against screening due to a lack of evidence supporting the practice. In order to receive a positive recommendation from the USPSTF, a screening program must be deemed both effective and economical. **Methods:** A literature review was conducted via the PubMed database through the Weill Cornell Medical Library Online to review screening practices for scoliosis. **Results:** A total of 43 articles were found; 20 articles were used for the literature review. **Conclusions:** Limited data exists to compare clinical outcomes of subjects screened for scoliosis versus non-screened. Data is even scarcer with regard to long-term follow up of subjects past adolescence. Because spinal malformations progress past adolescence in 68% of cases and at any given time over one-quarter of Americans have experienced low back pain in the past three months, it is important to provide the USPSTF adequate data to make an accurate recommendation regarding screening for scoliosis.

BIOGRAPHICAL SKETCH

Frank Jackson went to Rowan University and East Stroudsburg University. He worked at Moravian College in Bethlehem, PA as a member of the Sports Medicine staff prior to returning to school. Frank currently lives in Wood-Ridge, NJ with his wife and plans on moving to eastern Pennsylvania after graduating PA school.

Dedicated to dh.

ACKNOWLEDGMENTS

Thanks to my wife who has never stopped supporting me and is the reason I am still going. Thanks to all the family and friends along the way who have impacted me in some way, shape, or form.

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REVIEW OF THE LITERATURE

1.1 INTRODUCTION

Scoliosis is an abnormal lateral curvature of the spine that not only may produce a cosmetic deformity that can negatively affect one's psyche, but may also result in an increase in morbidity and mortality.[1-4] It has long been known that scoliosis causes a physical disability; however, 80% of patients have no identifiable etiology.[1] Since untreated deformities are known to progress, resulting in the possibility of eventual loss of physical ability, most physicians would agree that appropriate screening methods should be available to those individuals at greatest risk for abnormal curve progression, primarily adolescents.[1,2] Screening methods were introduced in the 1950s; however, due to the large number of unnecessary referrals and treatments, the United States Preventive Services Task Force advised against screening practices in 2004.[1] The topic remains an active controversy between governing bodies with groups such as the American Academy of Orthopedic Surgeons, the Scoliosis Research Society, the Pediatric Orthopedics Society of America, and the American Academy of Pediatrics all in support of screening in order to identify spinal deformities prior to progression.[1,3]

Idiopathic Scoliosis, named for its unknown etiology, is a complicated spinal deformity with lateral curvature and rotation of the vertebrae, resulting in what authors call a three-dimensional deviation of the spinal axis.[3,5] This abnormal curvature may cause a deformed and disfigured thorax shape, resulting in a convex and concave vertebral side which may inhibit rib movement and growth and, ultimately, thoracic expansion.[5] Asymmetries may also extend to structures beyond the musculoskeletal system, including viscera, fat and the skin, which are unique to each patient and progress along with the deformity itself.[6] While it is still unknown as to the exact causes of Idiopathic Scoliosis, it is clear that if left untreated it may result in increased morbidity and mortality.[2,7]

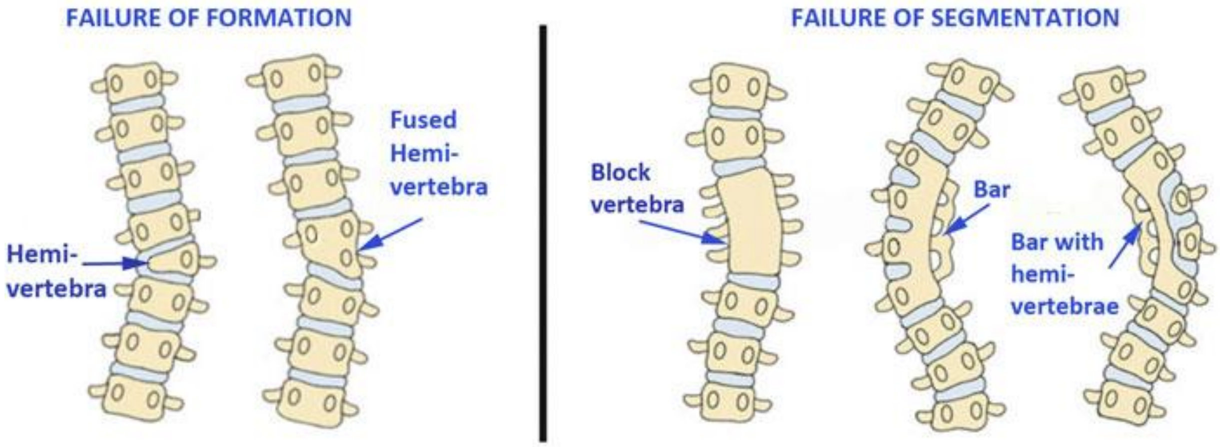
While idiopathic scoliosis includes abnormal lateral curvature of the spine that typically develops in the young patient and progresses throughout skeletal maturity, there are also deformities in the sagittal plane. When facing the patient, the spine has a normal convex and concave side, or lordosis and kyphosis, respectively. In order to keep the skull in a position to keep vestibulo-ocular equilibrium, the spinal column must be kept in appropriate alignment.[8] When one's kyphotic and/or lordotic curve is either exaggerated or lost, this balance must be achieved in other ways. For example, a patient with hyperkyphotic curve of the thoracic spine will eventually develop a hyperlordosis of the lumbar spine.[8] In order to appropriately balance the calvarium, the individual may also develop a hyperlordosis of the cervical spine depending on the amount of kyphosis of the thoracic region.[8] Another compensatory mechanism for abnormal sagittal alignment is pelvic rotation, or the lumbosacral relation.[8] In an individual with hypolordosis of the lumbar spine, the pelvis will posteriorly rotate around the hip axis, resulting in a sacral inclination that is more vertical.[8]

Several groups of idiopathic scoliosis exist, categorized here primarily according to their time of onset. The malformations relevant to this discussion typically present in adulthood, adolescence and childhood, and in infancy.[3] It is important to distinguish idiopathic scoliosis from congenital scoliosis, which is present at any age.[4,9]

Congenital scoliosis is a spinal deformity that results from a developmental abnormality, and while an abnormal curvature may not be present until later in life, some degree of vertebral malformation is always present at birth.[9] Up to 1 in 1000 live births will have some sort of congenital scoliosis, and there may be some genetic component to developing an abnormality.[9] Maternal exposure of toxins such as carbon monoxide, and maternal diabetes and use of antiepileptic drugs have been theorized to cause congenital scoliosis.[9] While a complete explanation of embryologic formation of the axial skeleton is out of the realm of this paper, it is relevant to understand the separate arrangement types of developmental failure.

Classification of developmental defects have been named according to the theorized malformation and their specific level (Fig. 1).[9] Three basic groups exist: formation failure, segmentation failure, and mixed types of malformation, which includes both segmentation and formation failures.[9,10] Formation abnormalities are graded according to the degree of malformation. Vertebrae may be wedge shaped and only partially deformed, with a hypoplastic side that creates height asymmetry.[9] Although the vertebral body may be misshapen, all of the parts of the vertebrae are present in this type of formation abnormality. In a more severe deformity of this etiology, a section of the vertebral body and a pedicle may be missing partially or completely, resulting in asymmetries of the vertebral column.[9] These formation failures can occur anywhere in the vertebral column and when adjacent to each other, may result in dramatic abnormal curvatures.[9]

Fig 1. Congenital Scoliosis. Reproduced with permission from the Scoliosis Research Society.



Segmentation failure is defined as an abnormal bony connection between adjacent vertebrae which can be unilateral, bilateral (termed a 'block' vertebrae), span several vertebrae on one side ('bar'), or be in combination with a formation failure (mixed malformation).[9] In congenital scoliosis, it is not uncommon to have associated rib abnormalities due to their close relationship with vertebrae during development. For example, certain types of formation malformations result in accessory and/or fused ribs, leading to further restrictions on spinal and thoracic growth which may result in severe functional consequences.[3, 4, 9]

After one rules out a congenital or other cause of the patient's malformation, idiopathic scoliosis becomes a diagnosis of exclusion.[3] While infantile idiopathic scoliosis is rare, rates among school-aged children are relatively high, with 1-2% of all children up to the age of 15 years suffering to some degree with spinal deformity of an unknown etiology.[3] Idiopathic scoliosis is the most common cause of lateral curvature of the spine and among the different categories of idiopathic scoliosis, Adolescent

Idiopathic Scoliosis (A.I.S.) remains the most common type that a clinician will encounter in practice, affecting up to 12% of the general population according to some researchers.[4]

It is well known that most of the population will experience back pain from various etiologies at some point in their life. In fact, 26.4% of Americans report low back pain lasting at least 24 hours in the past three months.[11] Knowing this, minimizing contribution from adult scoliosis becomes vital. It is estimated that scoliosis is present in 6% of individuals over the age of 50 and in 15% of patients with lower back pain over the age of 60 years.[12] In adolescent idiopathic scoliosis there is typically no presenting symptom; there is usually a clinical finding that leads to this diagnosis.[3] In adult idiopathic scoliosis, the patient will typically complain of back pain in the involved region, primarily the lumbar spine.[3] Lumbar degenerative scoliosis is one of the most common forms of spinal malformation in the adult population and, although it was previously believed that AIS was not a contributor to adult scoliosis, it is now known that curve progression can, and will, progress past the point of skeletal maturity and contribute to adult scoliosis, and thus chronic back pain.[4,12,13] Data show that spinal curves will progress beyond adolescence in 68% of cases, typically at the rate of 1° per year, potentially resulting in a significant deformity and disability.[4,12] In America, conditions that cause consistent pain affect 100 million adults and can cost upwards of $635 billion annually, so identifying conditions such as idiopathic scoliosis is important not only to the children and adolescents in whom it may develop, but also to adults who may benefit from a decrease in conditions that cause chronic pain and their associated costs.[11]

Since the majority of patients presenting in typical practice with spinal malformations are those with either adolescent idiopathic scoliosis or juvenile idiopathic scoliosis, the goal of this paper will be to discuss the screening methods of these conditions. Infantile scoliosis typically resolves on its own, with only 1/5 of cases persisting on to require treatment.[3] Although idiopathic scoliosis is typically an asymptomatic presentation as compared to its adult counterpart, identifying and addressing the malformation in youth before the age of skeletal maturity or curve progression can prevent morbidity and mortality, as well as chronic problems into and throughout adulthood.

1.2 METHODS

The online Weill Cornell Medical Library was utilized to retrieve all sources for the literature review, and the PubMed database was used exclusively. Search terms included 'scoliosis', 'scoliosis screening', 'screening' AND 'scoliosis', 'recommendations' AND 'scoliosis', and 'screening guidelines' AND 'scoliosis'. Articles were excluded based on their focus, included if they discussed scoliosis, its pathogenesis and sequelae, or current screening practices. The position statement regarding current screening recommendations from the AAOS was found utilizing the search terms 'American Academy of Orthopedic Surgeons scoliosis screening position statement' in the Google search database, which directed the author to the Scoliosis Research Society page of position statements.

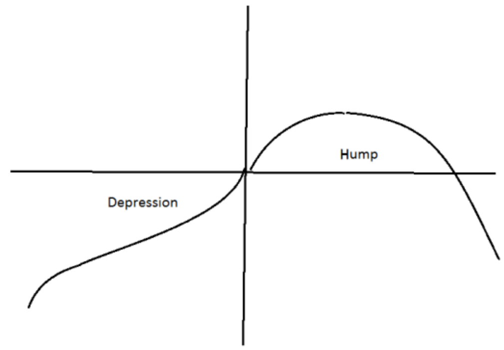
1.3 RESULTS

Forty-three articles were found for this literature review. Twenty-one articles were excluded because they did not adequately discuss scoliosis, its pathogenesis and sequela, or current screening practices. A total of 20 articles were used for the literature review, throughout all sections including the introduction, methods, results, discussion, and conclusion sections.

Determining whether scoliosis screening has any long-term benefit is a difficult task for a multitude of reasons. Reasons may include the length of study, ethical considerations regarding treatment, subject participation/ insufficient follow-up, clinician/researcher participation, and the great financial burden that a study like this may entail. While no studies currently exist that examine the impact of scoliosis screening and the results well into adulthood, there are very few studies that follow subjects until skeletal maturity.

A retrospective cohort study in Hong Kong conducted by Luk et al[1] examined school screening for AIS up until the age of 19 years in an attempt to determine the clinical effectiveness of school screening for AIS. The National Health Service began a program in 1995 that enrolled students in grades 5, 7, and 9 into a screening regimen. Students were examined initially by a trained physician or nurse using the Forward Bending Test and measured Angle of Trunk Rotation (ATR) using a scoliometer. The Forward Bending Test, moiré topography, measurement of the Angle of Trunk Rotation by Scoliometer, and rib hump measurement are the most commonly utilized screening tools, with Adam's Forward Bending test being the most

Fig 2: Example of a Humpogram



regularly used test due to its ease of use.[2,14,15] According to the Scoliosis Research Society/ American Academy of Orthopedic Surgeons position statement, the Forward Bending Test is the most specific for true scoliosis and should be included in all screenings, but one should not rely solely on it for the screening of scoliosis.[14,16] It involves the patient standing with feet together, knees straight, and bending forward at the waist.[17] From here, the clinician can evaluate the spine and identify any trunk asymmetries due to abnormal spinal curvatures and/or rib humps.[9,14,15] While performing the Forward Bend Test, humpograms (Fig. 2) can be drawn based on what the clinician visualizes and the angle of trunk rotation can be assessed using the scoliometer.[18,19] The scoliometer, also known as an inclinometer, is level-like tool that was developed to help deal improve objectivity of the Forward Bending Test.[14] The tool is placed flat on the patient's back and objectively measures trunk rotation away from the horizontal plane.[14,15,17] A study by Lee et al suggests using a cutoff of 15° to minimize unnecessary referrals.[14] Moiré topography is another relatively accurate screening method, yet due to its associated high costs, it is considered a second tier test.[14] moiré topography is a method of quantifying asymmetries based on the topography of the anatomy on the patients back examined by optical assessment then specialized photographs.[15]

In Luk's study, subjects were placed into different screening regimens depending on the ATR. If subjects had an ATR of 0-2°, they were screened biennially. Subjects with an ATR of 3° or 4° were screened annually. Students with an ATR between 5 and 14°, or evidence of trunk/shoulder asymmetry, were directly referred to Special Assessment Centres (SAC) for further evaluation utilizing moiré topography. Any child with an ATR >15° was sent directly to a specialty hospital that manages spinal deformities. If a Cobb angle over 20° was found then they would be followed until skeletal maturity. If the Cobb angle was under 20°, students would have repeated screening using moiré topography, ATR, and radiographs. Participants' medical records were traced until they turned 19.

The study revealed a prevalence of AIS of 2.49% for curves over 10°, 1.39% for curves over 20°, and 0.33% for curves requiring treatment. Of the screened students, 2.8% were sent for referral. Of the referred students, 43.6% had a Cobb angle over 20° and 9.4% required treatment for the abnormal curvature. The screening program was able to detect 88.1% of students with a Cobb angle over 20 degrees, and detected the need for scoliosis treatment in 4/5 (80%) of subject requiring intervention. The authors of this study recommended screening for scoliosis in a school setting similar to a program described here, as this program proved to limit the number of unnecessary referrals - the top reason agencies find screening programs to be not cost-effective.

In a second large-scale retrospective cohort in Hong Kong utilizing the same subjects as above, Lee et al[14] attempted to examine the current recommendations regarding selective screening for scoliosis. In an attempt to improve the accuracy of school-based screening programs and increase clinical effectiveness of the practice by limiting over-referrals, previous authors have suggested screening only the at-risk individuals. For example, since girls are more likely to develop AIS than boys, it has been proposed to only screen females.[14] In this study, researchers retrospectively examined the subjects from the study cited above but focused on the efficacy of screening selected individuals versus the results of the above study, as well as the value of 3 popular screening tests used.

As per the American Academy of Orthopaedic Surgeons (AAOP), female children should be screened both at age 10 and at age 12.[16] Boys should only be screened once between the ages of 13 and 14. According to the data collected by Lee, screening females as noted above would fail to identify, or identify too late for early intervention, 29.8% of girls with significant curvatures that develop after the age of 14. The number of males that would have late detected curvatures by following the AAOP guidelines was undetermined due to the screening ages in the study, but it is hypothesized that at least 3.9% of significant curves would be detected late. The American Academy of Pediatrics recommended screening both male and female children at ages 10, 12, 14, and 16.[16] There were 43 subjects that had significant curves detected via other sources that may have benefited from screening at age 16 or later. These 43 students, however, represented only 2.7% of all significant curves and proved that regular screenings this late into adolescence were excessive.

It is known that objective screening methods are recommended, and in the Hong Kong screening program, the forward bending test (FBT), the angle of trunk rotation (ATR), and moiré topography were utilized.[1] Research by Lee[14] proved that the use of moiré topography as a second tier test is highly sensitive, particularly when used in tandem with ATR with a 15° angle as a cutoff point. This is as opposed to direct referral to a specialist or by using ATR and/or FBT alone.

1.4 DISCUSSION

Screening programs to detect scoliosis in school-aged populations have long been implemented in many countries, including the U.S.[14] Continuance of this practice, however, has remained controversial due to the large number of unnecessary referrals for insignificant curves and debates around the cost-effectiveness of screening.[14,20] Since uninhibited abnormal curve progression has been shown to increase morbidity and mortality and decrease ones' quality of life, screening programs are implemented to intervene before the need for invasive surgical correction techniques becomes necessary.[1-4,14,21] While routine screening is recommended by a primary care provider at regular visit intervals, screening in a school setting, although controversial, is considered the most effective method and time in which to detect and intervene in curve progression.[18,19,21] It is known that some schools may already partake in routine screening for scoliosis, utilizing medical professionals such as nurses employed by the school district to perform evaluations and referring any abnormal findings to the child's primary care provider, thus increasing the potential for unnecessary referrals. Due to a lack of scientific studies regarding long-term follow-up of early intervention techniques, the recommendation by the USPSTF to support school-based scoliosis screening cannot currently be made.[15]

Many components must be considered when making a recommendation such as the one named above. For example, developing data would require a long-term study that may span over the length of a decade, until after the participants have reached skeletal maturity.[2] In order to evaluate long-term results such as prevention of chronic pain or scoliosis in adulthood, studies may be required to continue throughout the patient's lifetime, potentially requiring many different observers. Secondly, one must consider the adequacy of the screening methods.[19] The most commonly used and cost-effective forms of screening include the Adam's Forward Bending Test, use of the scoliometer, moiré topography, and rib hump measurement.[15] Of these, Adam's Forward Bending Test (FBT) is the most commonly used and, typically, the initial screening test of choice due to its ease of application.[2,14] The specificity of this test alone to identify scoliosis by visualizing a rib hump is 77.8%.[19] This leaves room for a large number of false positive results leading to unnecessary orthopedic referral potentially due in part to inexperienced clinicians and/or screeners.[14,19] Therefore it is recommended that this test be utilized along with other screening techniques in order to reduce the false-positive rate.[2] Finally, it is important to consider whether or not intervention in these patients can provide a favorable outcome.[19] Since the goal of early intervention is to provide conservative, non-invasive treatment options to prevent further complications, bracing is the standard accepted treatment option.[19] Bracing is intended to halt curve progression using external pressure via a hard plastic orthotic shell worn until skeletal maturity and requires much compliance on the patient's part.[22] As stated before, there is a lack of evidence investigating the long-term results of early intervention, which may include whether the implementation of bracing is effective at targeted intervention periods.[19,22] This author suggests the theory that earlier identification of spinal malformation may allow for an easier treatment regimen. Perhaps if a curvature is treated when it exists at a lesser angle it may prove to be more manageable and require less intervention. This may allow for bracing that is less of a disturbance to the patient, potentially increasing compliance with treatment and leading to more successful conservative outcomes.

Even after one considers existing scientific data surrounding the topic of school-based scoliosis screenings, cost-effectiveness of the screening program is a major consideration with regard to screening recommendations.[20] Screening has previously been considered cost-ineffective due to a high false-positive detection rate leading to a large number of referrals not requiring any intervention.[1, 20] A recent cohort of 115,190 students in Hong Kong examined the cost-effectiveness of screening students through the age of 19 and broke down its costs. According to the publication by Lee et al, the average total cost of screening a patient through the age of 19 by a trained physician or nurse was $17.94. When also considering diagnostic and medical care costs in the average cost of screening, the total cost were $20.02 and $54.63, respectively. For patients that required treatments such as bracing or surgical correction, respective 5-year costs of each were $8,018.85 and $27,538.97, with a minimum difference of $19,520.13 of surgery over conservative bracing. Since the early intervention initiated by screening programs is designed to reduce the number of surgical cases, one is forced to question the notion that school-based screening for scoliosis is not cost effective. Although it was noted that there was some overlap in patients requiring both bracing and surgery, it was determined that the screening would prove to be cost effective if up to 65% of the braced patients required surgery. A very small study from a Canadian Pediatric Hospital in Quebec suggested that up to 42% of the patients sent for suspected AIS were deemed ‘inappropriate referrals’, and 32% of patients being treated for AIS were considered ‘late referrals’.[19] This is echoed in the study by Lee et al that found that screening recommendations similar to those as suggested by the AAOS had an increased rate of late referrals, nullifying the goal of the screening program. In order to provide adequate and consistent data in order to support school-based screenings for scoliosis in the U.S., more long-term, large scale cohorts need to be done, with perhaps even longer follow-up times than those already completed.

The gold standard for evaluating spinal alignment is plain radiographs.[9] Screening each patient using radiographs is unrealistic due to several factors, including time, unnecessary radiation exposure, and financial constraints, so films are not used as a screening tool.[15] Patients with large curves are easy to identify and are more likely to be sent for further evaluation than a more subtle malalignment.[18] Therefore, screening tests designed to identify altered trunk shape and back deformities are used to determine the need for further diagnosis of scoliosis.[15]

The American Academy of Orthopedic Surgeons recommends screening at least twice for females, at ages 10 and 12 years old, and once for boys between the ages of 13 and 14 years old.[16] A retrospective cohort study conducted by Lee found that these guidelines have an increased rate of late referrals and/or missed curves that may be significant and require treatment.[14] In contrast, screening too early or often supports the notion that regular screening practices in schools prove to be clinically ineffective and not economically sound.[14] Finding a balanced protocol, perhaps similar to the Hong Kong screening program, with a tiered system that allows for some variability of measurement regimens and referral based on clinical findings is imperative if one wishes to construct a program that allows for early, potentially non-invasive intervention of significant, abnormal spinal curvature.[14]

1.5 CONCLUSION

School-based screening for scoliosis remains a controversial topic. The ultimate goal of screening has always been and remains to identify abnormal spinal curvatures and provide non-invasive intervention before they progress to the point where more aggressive treatments such as surgical correction becomes necessary in order to maintain quality of life. The United States Preventive Services Task Force currently recommends against the routine screening of children in the school setting, primarily because of a high number of unnecessary referrals, late presentation of the condition, and ultimately claiming that school-based screenings are not cost-effective enough to recommend continuance of the practice. In an attempt to provide a scoliosis screening practice that does not result in an over-abundance of unnecessary referrals, influential organizations suggested selective screening of children. The American Academy of Orthopaedic Surgeons recommended screening boys once, either at age 13 or at age 14. Girls, since they have a higher risk of developing an accentuated abnormal spinal curvature, should be screened twice; once at age 10, and again at age

12. The American Academy of Pediatrics recommends screening of both boys and girls at ages 10, 12, 14, and 16. The former method proved unsuccessful in accurately and effectively detecting abnormal curvatures prior to significant progression and the latter unnecessarily screened at an age deemed too late into adolescence to be economical.

Long-term studies examining the effectiveness of school based scoliosis screening remain close to non-existent; however, a large scale retrospective cohort in Hong Kong beginning in the mid-1990s proposed a screening program that has proved to be both successful in identifying significant spinal curves prior to the need for invasive intervention and cost-effective. More research must be available in support of the practice of school-based scoliosis screening that minimizes the number of false positives and ultimately improves its accuracy. Multiple studies recommend long term research, particularly retrospective cohorts and randomized controlled trials with sufficient long-term follow-up in order to analyze the enduring effects of scoliosis that may have been prevented or may have been recognized at an earlier point of development.

RESEARCH PROPOSAL

2.1 ABSTRACT

Problem: Insufficient data is available to make an adequate recommendation regarding childhood and adolescent scoliosis screening. The United States Preventive Services Task Force has reviewed existing data and current standards of the practice have been found to have an unnecessary number of false positive results which results in an avoidable amount of specialist referrals, ultimately proving the practice to be cost-ineffective. Current USPSTF guidelines recommend against the practice of screening of the aforementioned condition, contradictory to the recommendation of the American Academy of Orthopedic Surgeons, the Scoliosis Research Society, the Pediatric Orthopedics Society of America, and the American Academy of Pediatrics. **Purpose**: The purpose of this study is to determine the ability of a particular screening regimen to be successful in adequately identifying curvatures that may require treatment while maintaining cost-effectiveness by minimizing the amount of unnecessary referrals, ultimately improving the accuracy of school-based screening. **Research questions:** Is non-invasive screening for scoliosis in juvenile and adolescent patients associated with a significant decrease in abnormal curve progression in the teenage years and adulthood, decreased symptoms, and/or reduced morbidity and mortality? **Methods:** A large number of school-aged participants will be enrolled in a long-term randomized controlled trial to assess school-based screening practices and treated based upon a tiered referral system. Subjects will be monitored

biannually from young adolescence until the age of thirty. This will constitute the longest duration study on outcomes of scoliosis screening. **Outcomes:** Data will be collected throughout the study with the help of third-party data services. Subjects will be questioned on a number to topics related to the study, including back pain, disability, current/ past treatments and associated costs. **Benefit:** There is only limited data to evaluate clinical outcomes of scoliosis screening. Data with regard to long-term follow-up of subjects past adolescence is even scarcer. Because spinal malformations progress past adolescence in 68% of cases and at any given time over one quarter of Americans have experienced low back pain in the past three months, it is important to devise a protocol that improves the accuracy of scoliosis screening, minimizing the number of false positives and unnecessary specialist referrals.

2.2 AIMS

2.2.1 Project Overview

Schools that include students between the ages of ten and fourteen will be identified and, pending appropriated consent, will begin a regimen similar to that employed by Luk et al.[1] Unlike the previous study, however, these students will be followed well into adulthood to identify the outcomes of the program. Multiple aspects of the study will be tailored appropriately in order to minimize limitations and will be discussed further in the following paragraphs.

2.2.2 Research Question

Is non-invasive screening for scoliosis in juvenile and adolescent patients associated with a significant decrease in abnormal curve progression in the teenage years and adulthood, decreased symptoms, and/or reduced morbidity and mortality?

2.2.3 Specific Aims

AIM 1: To determine the number of patients diagnosed with new abnormal curvatures after a screening period (previously missed/undiagnosed).

AIM 2: To determine the number of patients requiring medical/therapeutic treatment related to spinal abnormalities in patients newly diagnosed with scoliosis.

AIM 3: To provide new data that may alter the current recommendations for scoliosis screening in youth and adolescents in order to provide better quality of life into adulthood.

2.2.4 Hypothesis

This author proposes that screening for scoliosis in adolescent patients will decrease the abnormal curve progression into patients' older years, decreasing complications later in life, including unwanted symptoms, morbidity and mortality, and will be cost-effective as a whole.

Alternately, screening for scoliosis will not prove to be significantly beneficial and will pose no benefit to patient symptoms and/or financial advantage.

2.3 BACKGROUND AND SIGNIFICANCE

2.3.1 Background

To date, very few studies have examined long-term outcomes of scoliosis screening. The USPTF previously had recommended scoliosis screening for children. However, due to an overall abundance of unnecessary referrals, the recommendation was reversed.

2.3.2 Project Significance

There is an ongoing debate between several of the involved governing bodies regarding the benefit screening for scoliosis in schools, including the American

Academy of Orthopedic Surgeons, the Scoliosis Research Society, the Pediatric Orthopedics Society of America, the American Academy of Pediatrics, and the United States Preventive Services Task Force. Those who recommend against screening in schools state that the practice is not cost-effective due in part to a high number of unnecessary referrals for further evaluation. While few studies have examined screening children and following them until skeletal maturity, no studies to date have followed individuals into adulthood while examining the effect of screening and ultimately the intervention of spinal deformities. Conversely, the effect of not screening and lack of intervention should also be examined, focusing on pain, risk of morbidity and mortality, quality of life, costs, and future complications.

Clinical screening for scoliosis is a fast and inexpensive method for recognizing possible problematic spinal curves. While it is understood that some schools do in fact employ screening methods that are typically performed by staff, particularly school nurses, researchers may be able to build off of these programs or initiate a regimen in schools that have no current screening guidelines in order to allow for a minimal amount of false positives and unnecessary specialist referrals. By identifying the long-term effectiveness of identifying such abnormalities as well as the effect of not identifying them, one could potentially reduce future complications and morbidity and mortality.

2.4 PRILIMINARY STUDIES

In a retrospective cohort study by Luk et al,[1] a total of 157,444 students were eligible for a biennial scoliosis screening, and their screening results and medical records up to 19 years of age were available. Students first had forward bending test and angle of trunk rotation (ATR) performed. Those with ATR between 5° and 14° or signs of adolescent idiopathic scoliosis were assessed by moiré topography regularly. Students with an ATR >15°, 2 moiré lines, or significant clinical signs were referred for radiography and had their Cobb angle measured. Of the 115,190 screened students in the cohort, 3228 (2.8%) were referred for radiography. At the final follow-up, the positive predictive values were 43.6% (41.8%–45.3%) for a Cobb angle 20° and 9.4% (8.4%–10.5%) for needing treatment, while the sensitivities were 88.1% (86.4%–89.6%) and 80.0% (75.6%–83.9%), respectively. This was the largest study that has demonstrated that school scoliosis screening in Hong Kong is predictive and sensitive with a low referral rate. Screening should thus be continued in order to facilitate early administration of conservative treatments.

2.5 RESEARCH DESIGN AND METHODS

2.5.1 Design

This study will be a longitudinal randomized clinical trial.

Schools in the United States that contain students between the ages of 10 and 14 will be identified regardless of their current participation in scoliosis screening. The study will utilize the Eastern and Western Seaboards due to their dense populations and ethnic variety. Schools already employing a screening regimen will be asked to adopt the program suggested by research team. After obtaining consent from school administrators, parents/guardians, and the participants, children will be split into 2 groups according to their school and location: one group will undergo school-based scoliosis screening, the other group will not undergo screening, with all subjects from a particular school assigned to a single randomized group. Children will be screened in a school setting and referred for further evaluation in a similar fashion to the study in Hong Kong by Luk.[1]

All participants included in the study will be followed from initiation into adulthood, monitoring them via questionnaire until the age of 30. Subjects will be asked about their symptoms and visits/referrals to physicians with regard to issues related to their back, particularly questions that correlate to spinal deformity, types of treatments, and costs involved.

2.5.2 Methods

Schools currently not undergoing screening for scoliosis and those that agree to adopt the study regiment will comprise the study. All schools in aforementioned regions of the US that contain children between the ages of 10 and 14 will be contacted and the school's representative(s) will be asked if they would be interested in participating in the study. For those that agree, consent must be obtained for participation, explaining the study and its purpose. A school and its children will be included if >100 of the children and their parents agree to partake in the study. A large number of subjects will be required in order to compensate for subjects lost to follow-up, as this study will go on for many years.

Because of its success in early identification and treatment of spinal deformity in the presence of cost-effectiveness, a screening method similar to the one proposed by Luk[1] will be employed. Subjects chosen for the screening group will undergo a school-based initial evaluation that utilizes the Forward Bend Test (FBT) and Angle of Trunk Rotation (ATR) led by clinicians chosen to monitor that subject's particular school. From there, subjects will be screened in a similar fashion every two years if they demonstrate ATR <2°. If the ATR is under 4 °, subjects will be screened annually. Patients with an ATR between 5° and 14° will then undergo moiré topography (MT). For individuals with less than 1 moiré line, they would then return to annual school-based ATR and FBT screening, with follow-up MT if ATR increases by 1° or more. For 1-<2 moiré lines, repeat ATR and MT are indicated every 6-12 months. For individuals with >2 moiré lines, posterioanterior x-ray of the spine to evaluate the Cobb angle is indicated. For patients with Cobb angle <20°, repeat ATR and MT every 6-12 months. For any patient with a Cobb angle >20°, or if ATR >15° is found at any point, referral by the supervising clinician for specialist evaluation is indicated. If a subject displays obvious signs of deformity, such as shoulder or trunk asymmetry, they shall be referred for further evaluation. Upon referral, all preceding medical information, including medical history and measurements previously obtained during the subject's participation in the study shall be sent with the subject in order to aid in further specialist evaluation. Figure 3 explains the screening regimen in a flowsheet format while figure 4 shows the timing of screening based on findings of tests.

Fig. 3: Screening Regimen

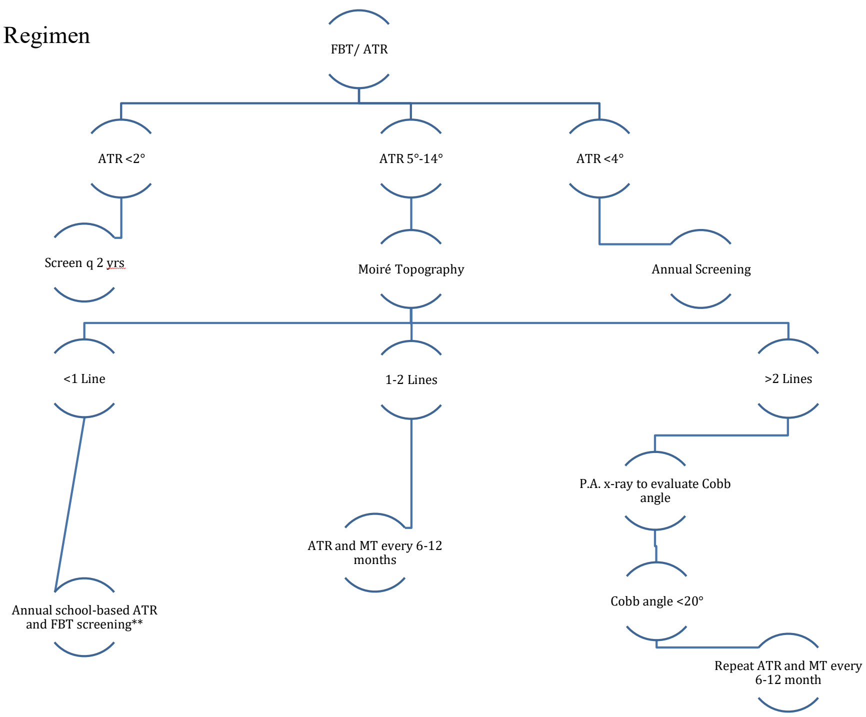
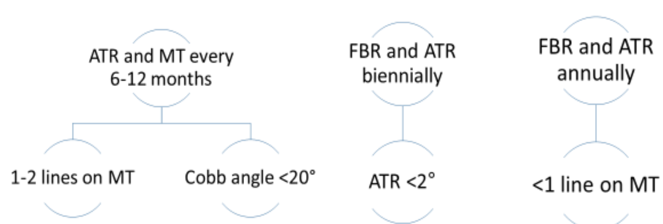


Fig. 4: Timing of screening according to findings



A medical questionnaire will be sent to all participants and their families every 6 months and will also include questions regarding updated contact information and preferred methods of contact, including mail, e-mail, phone, social media, or other new methods that may develop over the course of the study. Results of the questionnaire between the two groups will be evaluated by researchers and compared prospectively in order to evaluate the efficacy of this screening protocol with regard to identifying and preventing complications of spinal deformity throughout young adulthood.

The goal of this particular study is to determine if screening for scoliosis in youth as determined by the study's protocol (independent variable) has a significant effect on patient outcomes beyond skeletal maturity and into adulthood, including pain, increased risk of morbidity and mortality, quality of life, costs, and future complications (dependent variable).

The Scoliosis Research Society offers grants for research related to spinal pathology research, with idiopathic scoliosis, early treatment, and risk stratification being of particular importance. There are two application deadlines per year and both dates accept submissions related to the current topic. In 2015 there were 11 grants given, averaging $24,515 per award. Over the past 14 years, over $2.6 million has been granted for research related to spinal deformity. In order to help fund this study, researchers will apply for a grant from the Scoliosis Research Society.

2.5.3 Statistical Analysis

According to a priori power analysis, a minimum of 220 total participants are required, with two groups of 110 students. However, it is the belief of this author that many more participants should be utilized due to the length of the study and the potential loss of subjects. As stated earlier, 1-2% of children up to age 15 suffer from some degree of idiopathic spinal deformity.[3] By maintaining the minimum 220 participants as determined by the power analysis, researchers can expect only one or two children with deformity. In the data provided by Luk et al,[1] 157,444 children were identified for participation in the study. In order to provide an adequate number of subjects with an abnormal curvature, it is the belief of the author that at least 150,000 participant be identified to initiate the study. Similar to Luk's[1] study, children with a previous scoliosis diagnosis will not be screened and will not be considered to have been identified by the screening program. The remaining participants will be split into two equal groups and both sets will be examined: sample one is the control group and will not undergo school-based screening, while sample two will undergo the screening program. In each group, the number of subjects with an undesired outcome will be compared to individuals without a qualifying condition. Data will be analyzed via the chi-square test for association. Individuals meeting the following criteria will be identified and will be considered as an undesired outcome:

- Patients diagnosed with new abnormal curvatures after screening period (previously missed/undiagnosed)

- Patients requiring medical/therapeutic/surgical treatment related to spinal abnormalities in patients not previously diagnosed with scoliosis or not requiring treatment
- Patients experiencing new symptoms related to back pain not previously experienced

2.5.4 Limitations

As in any investigation, limitations are anticipated for this particular study. By attempting to predict potential problems we may be able to minimize future complication and increase the validity of the study. In order to locate an adequate number of subjects, the study should utilize heavily populated regions. This would allow researchers to find more participants without the need to travel to a new region. According to a 2012 United States Census Bureau News Release, the most heavily populated areas are the coastal states, particularly California and New York/ New Jersey. Locating schools along the coastal states should prove to be the most efficient strategy to find participants. Utilizing a large number of subjects may minimize the overall effect of subject loss.

To locate researchers and clinicians, academic institutions in each particular region will be contacted. Interested individuals will be responsible for an area and screening subjects and selected schools in that area. All levels of clinicians may be utilized, pending proper instruction and scheduled training workshops. If interested in participating in the study, school nurses may also partake in screening, but should be included in procedural and scheduled clinical training for all clinicians. If clinicians are lost during the study, as they graduate residency, move, change positions, or for any other reason, new individuals will be recruited. If programs are interested, participating may be built into part of a residency or fellowship program.

To ensure uniform screening procedure by the clinicians, any practitioner who will partake in the screening process will undergo formal training and evaluation. At the initiation of the program, researchers shall go to each of the regions where clinicians will be performing the screening procedures to hold an informative lecture and workshop to educate the clinicians on proper procedure and analysis. Prior to beginning the screening, clinicians' capacity to participate in the program will be evaluated via multiple choice test and re-evaluated annually. For any individuals who may later enter the program, the possibility of an online lecture may be considered in lieu of a formal in-person workshop.

Results of each screening session shall be documented by the clinicians assigned to each particular school and/or region. As previously stated, questionnaires will be sent to the families of participants every 6 months based off of the initial screening, and the individual's data will be updated according to their response. In order to ease the burden of organizing and sending individual questionnaires, 3rd party printing and mailing services such as Vistaprint® will be utilized. Similarly, data collection will be provided by medical data collection services such as Tonic®. Third party services may be utilized after appropriate consent is obtained from participants.

Once it has been determined that an individual must be referred for imaging as per the protocol, the participant will be sent to a radiology group that utilizes digital formatting of images. These images will be sent to and read by a 3rd party radiology service that has the capability of measuring and evaluating Cobb angles. This data will then be collected by the clinicians responsible for that individual and screening will carry on as per the protocol and the result of the Cobb angle.

One may consider it unethical to withhold treatment/screening; however, we can minimize this thought by utilizing populations that do not currently undergo school-based screening. The current recommendations are against screening for scoliosis in schools, so by providing a school-based screening regimen, researchers will either be providing equal or increased amount of care than currently received. Also, any time one uses children as study participants, one must pay particular caution as children are vulnerable subjects. Either one or both parents must provide consent for participation depending on the level of risk, as determined by the Institutional Review Board.[23] If it is determined that the study proposes 'minimal risk', defined as 'probability and magnitude of harm or discomfort anticipated in the research are not greater in and of themselves than those ordinarily encountered in daily life or during the performance of routine physical or psychological exams or tests', then only one parent is required for consent. It is hypothesized that the proposed study may prove to be more than minimal risk for a number of reasons. Primarily, and most obviously, is the risk of radiation exposure during Cobb angle evaluation. While this is limited only to subject who require referral for the aforementioned evaluation, radiation exposure risk is present any time a patient has x-rays performed. Secondly, researchers must be aware of the potential for subjects to feel segregated by peers when the need for further evaluation is warranted. While clinicians and researcher will attempt to minimize this psychological adverse effect by adhering to patient privacy laws and regulations regarding confidentiality, the potential still exists. Finally, potential for parental exploitation for monetary gain must be realized and minimized. Efforts to address this will be discussed further. After level of risk is determined by the IRD, researchers will obtain the appropriate consent for participation in the study as deemed necessary. Any individuals that are interested in obtaining guidance regarding the level of risk of a particular study that utilizes human subjects, an application may be filed with the Institutional Review Board on the American Public University System website.

While children themselves cannot give consent, it is important to include them in the consent process.[23] Age appropriate information should be provided to each subject and time allowed for them to determine whether they would like to participate in the study.[23] The child's decision for assent or dissent should be honored and the ability to withdraw from participation be upheld at all times.[23] It has been determined that children over the age of eleven have a higher capacity to understand the elements of a study than their younger counterparts, so it is theorized that the current study's utilization of subjects at an initial age of 10-14 year olds should prove to be successful in allowing for adequate and ethical participation.[23]

In addition to risk evaluation, rewards for participation should be appropriate.[23] For example, one should avoid large sums of cash as it may provide the potential for parents to exploit children for monetary gain.[23] Reimbursement for travel, meals, time-off from work, as well as more age-appropriate gifts for subjects such as movie ticket vouchers may be considered.[23]

2.5.5 Timeline

Students will undergo initial screening if they are between the ages of 10 and 14. The Hong Kong study by Luk[1] screened and examined patients through the age of 19, currently the longest study to examine the effects of school-based screening. The current study aims to monitor subjects into adulthood in order to adequately determine the effect of childhood and adolescent school-based screening for scoliosis on symptoms commonly found in adults, particular back pain and the impact on one's quality of life. In order to determine this impact, subjects must be followed beyond any previous study. This author proposes following patients through the following decade until the age of 30, continually monitoring symptoms in order to determine the effects on the adult subjects.[23]

2.5.6 Summary

This study design is intended to provide a better understanding of the benefits of screening for scoliosis in youth and adolescence. By following study participants through adolescence and into adulthood, researchers may be able to identify a reducible risk of preventable morbidity and mortality in adulthood associated with unidentified or untreated spinal curvatures. In order to minimize unnecessary referrals, a tiered referral system similar to the one proposed by Luk will be utilized. By preemptively identifying research limitations, researchers will hopefully be able to increase the success rate of the study.

2.6 CONCLUSION

The United States Preventive Services Task Force develops recommendations with regard to screening practices, counseling, and preventative medications. In an attempt to improve the overall health of US citizens, the members of the USPSTF assess peer-reviewed evidence and recommendations are made according to existing data. Each recommendation is organized according to its strength, and assigned a letter grade for the quality of research and the USPSTF's level of support. As of 2004, screening for scoliosis in adolescents has been assigned the letter grade 'D', which indicates that there is moderate to high certainty that the service has no net benefit or that the harms outweigh the benefits, ultimately resulting in a recommendation against the practice. The USPSTF is now reviewing current data in anticipation for a topic update scheduled for release in 2017.

Currently, there is an unacceptably small amount of research on the topic of screening for scoliosis. The USPSTF does an excellent job of providing evidence-based recommendations determined by the current data available; however, there is not enough evidence to make an accurate recommendation to endorse the practice. In order to provide an appropriate recommendation, there must be more accurate data to support or refute the long-term effects of scoliosis screening during adolescence. While some data exists to evaluate the impact of screening on immediate outcomes, particularly regarding the need for surgical intervention or bracing, an extremely limited amount of research is available to follow participants through a longer period to determine its efficacy over time. By employing a staged referral system, the chances for an unnecessary referral and ultimately treatment are minimized, resulting in a program that is both cost-effective and efficacious.

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