Metrology of Health-Related Quality of Life Measurements in Adults and Adolescents with Scoliosis

by

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Abstract

***Background*:** Scoliosis impacts Quality of Life (QOL). Stabilization exercises have been shown to be effective for improving patient reported outcomes (PROMS.); however, our literature review examining the effects of such exercises on QOL showed that such studies in adults with scoliosis are lacking. PROMs with adequate measurements are prerequisites for conducting such studies. However, there has been no head to head comparisons to identify the best tools for measuring changes in QOL in the same samples of adults and adolescents with scoliosis. Widely used QOL PROMs have limitations such as ceiling effects, and insufficient reliability and validity evidence. Recently developed quality of life PROMs including the Italian Spine Youth Quality of Life (ISYQOL), Truncal Anterior Asymmetry Scoliosis Questionnaire (TAASQ), and Body Image Disturbance Questionnaire-Scoliosis (BIDQ-S) aim to address these limitations but a thorough metrological comparison is needed.

Objectives: This thesis aimed to: Systematically review the effects of stabilization exercises on pain, disability, and quality of life in adults with scoliosis, compare the test-retest reliability and convergent validity of the two versions of the Spinal Appearance Questionnaire (SAQ) in Adolescents with Idiopathic Scoliosis, and determine the test-retest reliability and convergent validity of the BIDQ-S, TAASQ, and ISYQOL scores in adolescents and adults with scoliosis against radiographic measurements and established quality of life questionnaires (Scoliosis Research Society 22r [SRS-22r] and SAQ v1.1).

Methods: First, a systematic review on stabilization exercises for adult scoliosis was conducted on March 9, 2017 of Medline, CINAHL, Embase, SportDiscus and the Cochrane Register of Controlled Trials. Data extracted included information about participants, treatments, and results on pain, function and quality of life. Each study was appraised for quality using the Cochrane Risk of Bias tool.

For the 3 metrological studies, two established questionnaires (the Scoliosis Research Society-22 refined (SRS-22r) and the Spinal Appearance Questionnaire (SAQv.1.1)) and three new questionnaires (ISYQOL, TAASQ, and BIDQ-S) were collected along with radiographic measurements (Maximum Cobb angle, coronal balance, and thoracic and lumbar vertebral rotations). Questionnaires were administered electronically twice with a one-week interval between.

Results: The systematic review found limited evidence from only one study with high risk of bias that stabilization exercises significantly improve pain, disability and quality of life.

The test-retest reliability of the SAQ domains ($ICC_{3,1}$ = 0.72 to 0.94) in adolescents with scoliosis was similar or slightly lower than for the SAQ v1.1 Total, Appearance, and Expectations domain scores ($ICC_{3,1}$ =0.86 to 0.94). Significant correlation was observed between the SAQ scores and both the SRS-22r Total score (r= -0.35 to -0.59) and the Cobb angle (r= 0.38 to 0.59). The SAQ v1.1 Total score correlated with the SRS-22r Total score (r=-0.50) and with Cobb angle r = 0.35 to 0.63. The ceiling and floor effect analysis favored the newer SAQ v.1.1.

Test-retest reliability estimate ($ICC_{3,1}$) for new questionnaires in adolescents with idiopathic scoliosis ranged from 0.77 to 0.95 compared to 0.70 to 0.93 in adults with scoliosis. Correlations that met our threshold and supported convergent validity between the new and established questionnaires numbered in adolescents with scoliosis and ranged from r = 0.35 to 0.70 compared to r = 0.38 to 0.72 in adults with scoliosis. Correlation between radiographic measurements and new questionnaires that met our threshold and supported convergent validity ranged from r = -0.35 to -0.47 in adolescents with scoliosis compared to r = -0.35 to -0.60 in adults with scoliosis.

In adolescents with scoliosis, only the ISYQOL did not present notable ceiling or floor effects (≥15%). Both, the ISYQOL and the BIDQ-S did not present notable ceiling or floor effects (≥15%) in adults with scoliosis.

***Conclusion*:** This thesis highlighted the need for studies evaluating the effects of exercise on QOL in adults with scoliosis. In addition, it suggested that clinicians and researchers should use the SAQ v1.1 instead of the old version of the SAQ. In general, all new questionnaires demonstrated convergent validity, adequate reliability and acceptable ceiling and floor effects levels for research use in both adults and adolescents with scoliosis.

PREFACE

This thesis contains original work by Malik Alanazi completed under the supervision of Eric C. Parent, Associate Professor in the Department of Physical Therapy at the University of Alberta; Douglas P. Gross, Professor in the Department of Physical Therapy at the University of Alberta; and Josette Bettany-Saltikov, Senior Lecturer at Teesside University, United Kingdom. Studies in Chapters 3, 4, and 5 were approved by the University of Alberta Health Research Ethics Board (Pro00073569). **(Appendix 1.1)** I was the primary author of all the chapters in the thesis but received input on my manuscripts from co-authors and from my supervisory committee members for all chapters.

The systematic review in chapter 2 was published in the European Journal of Physical and Rehabilitation Medicine in 2017. (Alanazi MH, Parent EC, Dennett E. Effect of stabilization exercise on back pain, disability, and quality of life in adults with scoliosis: a systematic review. Eur J Phys Rehabil Med. 2018;54(5):647-653. doi:10.23736/S1973-9087.17.05062-6)

My role in this chapter was conceptualizing the idea, screening all abstracts and full texts, extracting data, quality appraisal, and writing the manuscript. I received assistance from Dr. Eric C. Parent in screening and data extraction. Elizabeth Dennett helped in preparing the search strategy. Alex Su assisted in the screening process as the second reviewer.

In Chapters 3, 4, and 5, I contributed to the conceptualization of the idea with Andrea Lin. I assisted Dr. Eric C. Parent in data collection and scoring strategy. Dr. Jim Mahood, Dr. Eric Huang, Dr. Marc Moreau, Ms. Sarah Southon and Ms. Kathleen Shearer contributed to the recruitment of

adolescents. Dr. Jim Mahood, Dr. Babak Sharifi, Dr. Eric Huang and Sanja Schreiber contributed to recruitment process of adults. Additionally, I was responsible for data analysis and writing the manuscripts for all chapters with input from Dr. Eric C. Parent. I did the image analysis with assistance from Kenwick Ng and Mathew Shaker. I also received feedback from Prof. Douglas Gross and Dr. Josette Bettany-Saltikov on the planning and the reporting work for these chapters. Dr. Jim Mahood, Dr. Eric Huang, Dr. Marc Moreau, Ms. Sarah Southon, Ms. Kathleen Shearer and Andrea Lin were given an opportunity to provide editing input for the adolescent manuscript. Dr. Jim Mahood, Dr. Babak Sharifi, Dr. Eric Huang, Dr. Sanja Schreiber, and Andrea Lin were given an opportunity to provide editing input to the adult manuscript.

For Hamoud and Radhwa, my precious parents.

For Munirah, the greatest wife in my eyes.

For Norah, Tamem, Jassar, and Sarah my kids.

...with all my love

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1 CHAPTER 1– INTRODUCTION

Historically, scoliosis was defined simply as an abnormal, lateral curvature of the spine.[1] However, other integral factors are now recognized as being associated with this lateral (or coronal) curvature, namely a loss of sagittal balance and malrotation of the spine.[1] Therefore, scoliosis has been redefined as "a complex three-dimensional rotational deformity that affects the spine in the coronal, sagittal, and axial planes."[1] A patient is diagnosed with scoliosis when the Cobb angle of the lateral curvature exceeds 10° on a standing radiograph in the presence of vertebral rotation.[2] The Cobb angle is a widely used and accepted measurement to evaluate scoliotic curve severity and risk for progression.[3]

Scoliosis significantly impacts Quality of Life (QOL) in affected patients (refer to Section 1.4.5).[4,5] Scoliosis management includes operative and non-operative interventions.[4,6] Surgical interventions are associated with high rates of complications especially in older patients.[4] Thus, surgeons prefer to use conservative management such as exercise to initially manage the symptoms of scoliosis.[4] Despite this, there is limited published evidence supporting the effects of exercise on adult patients with scoliosis. To examine the effects of exercise on patients with scoliosis, outcome measurements that adequately characterize the effects of treatment are necessary. This PhD thesis aims to assess the measurement properties of three recently developed, Patient Reported Outcome Measures (PROMs) which were developed to address limitations of currently used questionnaires in the adolescent population. Since there are currently no PROMs developed specifically for the adult population, this thesis seeks to examine how these PROMs (the Italian Spine Youth Quality

of Life [(ISYQO), Truncal Anterior Asymmetry Scoliosis Questionnaire [(TAASQ], and Body Image Disturbance Questionnaire-Scoliosis [BIDQ-S]) perform in adults with scoliosis.

This chapter provides information about scoliosis along with the different types of scoliosis, etiology, prevalence, signs, symptoms, and management approaches. Furthermore, outcome measures including the limitations of the currently used outcome measures (SRS-22r and SAQ) will be discussed.

1.1 Types of Scoliosis

Scoliosis can be classified according to the time of onset, ranging from infantile (<3 years), to juvenile (3-10 years), to adolescent (10 -18 years), and to adult (>18 years) scoliosis[7]. Scoliosis can also be classified according to etiology or cause, such as congenital (due to birth deficits), neuromuscular (due to neuromuscular diseases such as cerebral palsy), and idiopathic (unknown etiology).[8] The focus of this project will be mainly on adult scoliosis (degenerative or idiopathic) and adolescent idiopathic scoliosis (AIS), as they make up the most prevalent types of scoliosis.[9–11]

Adult scoliosis can be categorized into two major types: adult degenerative (also called de novo scoliosis) and adult idiopathic scoliosis.[12] Adult degenerative scoliosis (ADS) is "a condition in which the lumbar scoliotic curve typically develops after the age of 50 in patients who did not have childhood scoliosis."[13] In contrast, adult idiopathic scoliosis is a continuation of untreated or residual adolescent idiopathic scoliosis into adulthood.[12,14] Adolescent idiopathic scoliosis (AIS)

is defined as “a structural, lateral, rotated curvature of the spine that arises in otherwise healthy children at or around puberty”.[15]

1.2 Etiology

Despite decades of research, the etiology of adult and adolescent idiopathic scoliosis remains unknown. It has been suggested that scoliosis is a “complex genetic disorder where genetic factors interact with patients’ environmental factors and growth to create a spinal deformity.”[2] ADS is a result of collective degenerative changes that occur progressively over a person’s lifetime.[16] As a person ages, the intervertebral discs experience dehydration, asymmetrical degeneration, and collapse.[16] This usually occurs in addition to facet degeneration and ligamentous laxity.[16] These degenerative processes may have synergistic effects on each other, leading to asymmetric loss of disc height. [17,18] This consequently causes the development of axial rotation and coronal imbalance that results in ADS.[17,18] ADS can also develop as a secondary condition stemming from leg length discrepancy, hip pathology, or metabolic bone diseases (such as osteoporosis).[14]

1.3 Prevalence

Adolescent idiopathic scoliosis is the most common form of scoliosis among children.[19] The prevalence of adolescent idiopathic scoliosis has been estimated to be between 2-3%[20] and represents 70 to 80% of all adolescent spine deformity cases.[21] In North America, adult idiopathic scoliosis is the least prevalent type of adult scoliosis, estimated to make up 0.4–3.9% of all cases.[22] Detailed information about the prevalence of adult idiopathic scoliosis is not available within the current literature. However, information about the prevalence of adolescent idiopathic scoliosis

may help understand the prevalence of adult idiopathic scoliosis since the latter is a continuation of the former. The ratio of females to males is almost equal (1.3:1) when the curve magnitude is 10-20°, but for curves of 20-30° magnitude, the ratio increases to 5.4:1. Lastly, for curves above 30°, the ratio of women affected to men affected is 7:1.[23]

In an American study, the prevalence of adult degenerative scoliosis (ADS) was estimated to be as high as 68% for people over the age of 60, having an average age of 70.5 years.[9,10] The lowest prevalence estimation reveals that at least 5.88 million adults in the United States experience adult scoliosis.[9,10] ADS is the most common form of scoliosis observed in middle-aged and older adults.[24] The female to male ratio of those with ADS is relatively equal.[14] In adults with ADS, the prevalence is inversely proportional to curve magnitude. For example, the rate of curves measuring 10°, 10–20°, and >20° are 64%, 44%, and 24%, respectively.[25]

The risk of developing health problems secondary to scoliosis for adults includes decreased quality of life, cosmetic deformity, pain, and progressive functional limitations if the curve magnitude is 30° or more at skeletal maturity.[26] If the curve is over 50°, it is almost certain that the curve will progress into and throughout adulthood, causing QoL limitations and other health issues related to neurological and cardiopulmonary function.[26]

1.4 Signs and Symptoms

1.4.1 Curves

In patients with adolescent idiopathic scoliosis, right thoracic curves represent around 80% of all cases.[17] In skeletally mature adolescents, curves < 30° are unlikely to progress, whereas thoracic curves > 50° may progress up to 0.5-1° per year.

Patients with ADS tend to have smaller Cobb angles compared to patients with adult idiopathic scoliosis.[4] The rate of curve progression in ADS has been reported to be 1-6° per year, with an average of 3°.[17] In contrast, curve progression in adult idiopathic scoliosis is slow, reportedly 0.5° per year.[17] While rotational and lateral components generally involve only the apical vertebrae in ADS, curves in adult idiopathic scoliosis typically involve a multi-level rotational component and lateral listhesis (defined as a lateral translation of the vertebral body towards the convexity of the curve).[4,27] Curves in ADS are typically located in the lumbar region with an apex at L3, whereas curves in patients with adult idiopathic scoliosis can have thoracic, thoracolumbar, or lumbar curves.[4]

1.4.2 Pain

Lower back pain is prominent in patients with ADS and prevalent in roughly 60 to 93% of cases. This is higher than the 1-month prevalence of lower back pain in the general population of adults, which is estimated to be 23.2%.[28,29] Back pain is a less common complaint in people with adolescent or adult idiopathic scoliosis, and it is usually associated with larger curves or radicular leg pain.[4] Generally, back pain in adults with scoliosis results from spinal imbalance (coronal or sagittal imbalance) or spinal stenosis (central or foraminal stenosis).[30]

1.4.3 Neurogenic Claudication

One of the most important symptoms in patients with ADS (not common in patients with idiopathic scoliosis) is neurogenic claudication, which is caused by central spinal stenosis and presents as pain in the lower back as well as numbness and weakness in one or both legs.[4] Spinal stenosis is defined as a pathological condition in which the neural elements of the spine are compressed by bone and soft tissue which obstructs blood flow, resulting in ischemic pain.[31] Spinal stenosis is more common in adults with ADS (90%) as opposed to adults with adult idiopathic scoliosis (31%).[32] The symptoms of spinal stenosis are similar in both types of adult scoliosis.[31] Due to central stenosis, patients with neurogenic claudication mainly complain of bilateral leg weakness and pain when walking or standing; this pain reportedly improves with sitting or bending forward.[33]

1.4.4 Radicular Symptoms

Smith and colleagues[34] reported that 64% of patients with ADS seeking operative treatment have severe radicular leg pain. Radicular pain is a combination of back and leg pain, with pain being worse below the knee in the affected leg. It is thought to be caused by foraminal (lateral) stenosis.[4] Although radicular symptoms naturally occur on the concave side of the curve, stretching of a nerve on the convex side may also produce radiculopathy.[4] Radicular symptoms are not usually an issue for patients with idiopathic scoliosis.[4]

1.4.5 Quality of Life

Scoliosis does not affect QOL in adults and adolescents with scoliosis in the same manner. ADS significantly impacts functional ability due to the severe back and leg pain it causes.[4] Furthermore, the risk of pain, functional impairment, and disability increases with age. Among adult patients, occupational impairment has been noted leading to an increased incidence of sick leave.[35] Therefore, patients with adult scoliosis typically seek surgery to relieve symptoms and to improve function.[5] Still, appearance issues may drive adults with scoliosis to undergo surgery. Scoliosis involves malalignment of the spine in the coronal, axial, and sagittal planes.[4] The curve magnitude (measured with the Cobb angle) in the coronal plane does not seem to have a significant impact on function in patients with ADS.[36] In contrast, the activity domain of the SRS-23 patient questionnaire strongly correlates with malalignment in the sagittal plane (SVA) ($r = -0.49$).[36]

In contrast, patients with adolescent idiopathic scoliosis are often asymptomatic.[5] The presence of appearance-related problems during adolescence is thought to be of greater influence on perceived appearance than in childhood or adulthood. Further, it is more common among adolescent females due to feelings of unattractiveness, expressed by difficulty finding clothes and dissatisfaction with appearance.[37,38] Other effects of adolescent idiopathic scoliosis include depression and poor sense of self-image resulting from the change in appearance caused by scoliosis. [39] Typically, therefore, adolescents with scoliosis undergo surgery to improve cosmetics and minimize future curve progression, thereby improving patient self-confidence.[5]

Eventually, the overall QOL in both populations is significantly affected by the disease. Therefore, having appropriate QOL outcome measurement tools is necessary for accurate evaluation and treatment.

1.5 **Treatment Approaches**

1.5.1 Non-operative Management

Therapeutic Exercise

Exercise is one of the main treatment options used by physical therapists.[40] However, the use of exercise to treat scoliosis is controversial.[6] Most physical therapy centers in the United Kingdom and the United States do not advocate for its use in patients with scoliosis, despite the fact that it is routinely used in France, Germany, Italy, and many other countries in continental Europe.[6]

Physiotherapeutic Scoliosis-Specific Exercises (PSSE) are recommended by the Society of Scoliosis Orthopaedic and Rehabilitation Treatment (SOSORT) as a first line intervention to treat adolescent idiopathic scoliosis.[26] The main elements of PSSE are: (1) 3D auto-correction, (2) stabilization of the corrected posture, and (3) education of the patient and family about the effects of scoliosis and impact made by exercise on the posture of the spine.[26] Some PSSE rely on self-correction movements that are performed to reach the best possible realignment of scoliosis curves with the help of external tools.[41] Examples of approaches following this school of scoliosis physical therapy are the Schroth Method, Barcelona Scoliosis Physical Therapy School (BSPTS), DoboMed, Side Shift, and the Functional Individual Therapy of Scoliosis (FITS).[41] Other PSSE

approaches follow a different school, called Scientific Exercises Approach to Scoliosis (SEAS).[41] SEAS differentiates self-correction and exercise; patients are first taught the best possible alignment of the spine and then perform exercises or movements to maintain that position.[41]

The Schroth Method is one of the most common SSE approaches.[42] It consists of scoliosis-specific sensorimotor, postural, and breathing exercises.[42] A fundamental component of the Schroth Method is auto-correction, or attempts by the patient to reduce spinal deformity by active postural realignment of the spine in three dimensions. Auto-correction is achieved through self-elongation and postural corrections specific to each curve pattern, and is eventually integrated into daily activities.[42] The effect of Schroth exercises on adolescent scoliosis has been investigated in studies that have shown some promising results.[43] However, no randomized controlled studies have been conducted to evaluate the effect of Schroth exercises on adults with scoliosis.

Another intervention broadly used by physical therapists to treat the spine is core-strengthening exercises. Core exercises are defined as "the restoration or augmentation of the ability of the neuromuscular system to control and protect the spine from injury or re-injury." It is used to describe a spectrum of exercises that have the common goal of improving lumbopelvic control such as plank and side and bridge supine[44,45]

Core stabilization exercises have been shown to be effective for improving pain and biomechanical function in the lower backs of patients.[28,46,47,48] However, current literature lacks evidence to support the positive effects of these exercises on adults with scoliosis.

Brace Treatment

In general, braces can be soft, rigid, super-rigid, or made of plaster cast. Using braces in skeletally-immature patients with adolescent idiopathic scoliosis is a well-accepted treatment to prevent curve progression.[49] Palazzo and colleagues[50] assessed the effectiveness of underarm bracing in 38 adults with degenerative scoliosis and found that the progression rate decreased from 1.47° ± 0.83°/year without bracing to 0.24° ± 0.43° /year with bracing ($P < 0.0001$). For idiopathic scoliosis, an average progression rate of 0.7° ±0.06°/year was measured without the use of braces, and this progression rate changed to 0.24° ±0.43° /year with the use of bracing ($P = 0.003$). The mean follow-up time was 8.7 ± 3.3 years after bracing.

De Mauroy and colleagues[51] conducted a study to investigate the effect of the Lyon management treatment which includes the use of a rigid brace in association with specific physiotherapy. The study included 158 patients with adult scoliosis who were followed at least for 5 years. [51] The study concluded that the Cobb angle was stable in 56% of the participants, improved more than 5° in 24% of the participants, and worsened more than 5° in 20% of the participants.[51] However, the type of scoliosis each patient had and type of physiotherapy used was not specified in this study.

1.5.2 Operative Management

The main goal of surgical intervention in adults with scoliosis is to decrease pain and disability, thereby allowing patients to have an acceptable level of physical function.[4] Indications for surgery include: (1) patients who did not improve with non-operative interventions in symptomatic relief or functional recovery; (2) patients whose symptomatic lumbar curves have coronal Cobb angles higher than 30°-40°; and (3) patients with significant curve progression (>10° per year).[22]

Surgical management of adult deformities such as scoliosis may include a variety of operative approaches, ranging between limited interventions (such as decompression of the neural elements without fusion) to extensive surgeries for realignment of the spine.[52] Due to the high complication rates associated with surgical care and poor bone quality (especially in older patients), surgeons prefer to use conservative management as a starting point to treat the symptoms of scoliosis.[4]

1.6 **Outcome Measures**

The term '*patient-reported outcome measure' (PROM)* refers to "any assessment coming directly from patients, without interpretation by physicians or others, about how they function or feel in relation to their health condition."[14,53] PROMs play an important role in research and clinical care[14]. In research, PROMs play a fundamental role in facilitating the comparison of intervention outcomes. In clinical care, they are used to demonstrate the overall effectiveness of different treatment approaches. Furthermore, PROMs help healthcare providers communicate with their patients, helping patients to identify their own expectations and outcomes of treatment.[14,54]

PROMs may identify: 1) disease symptoms or treatment side effects, such as pain, fatigue, or anxiety; 2) functional outcomes, such as changes to physical, sexual, social, emotional, or cognitive function; or 3) multidimensional constructs.[55] The majority of PROMs used and discussed within scoliosis literature are health-related quality of life (HRQOL) measures.[56] Health-related quality of life is an assessment of the effects of a disease or a treatment on the physical, psychological, and social domains of functioning and well-being for a patient.[55] In scientific

literature, HRQOL PROMs are divided into generic instruments, disease-specific instruments, and super-specific instruments.[14,56,57]

1.6.1 Generic Instruments

General or generic instruments are questionnaires that are designed to evaluate health-related quality of life in the general population. The most common examples are the Medical Outcomes Short Form 36 (SF-36) and the EuroQol 5-Dimension Questionnaire (EQ-5D).[56,57]

Medical Outcomes Short Form 36 (SF-36)

One of the most widely-used, generic HRQOL instruments is the SF-36, which has been translated into over 40 languages and aims to assess a patient's general condition.[14,58] The SF-36 contains 36 questions related to eight different domains: physical functioning, bodily pain, social functioning, mental health, vitality, role limitations due to physical health, role limitations due to emotional problems, and general health perceptions.[4] In addition, the SF-36 includes a single item that focuses on the perceived change in general health status over a one-year period (health transition).[4] The SF-36 can be completed and scored using a 0-100 scale, where 0 indicates the worst possible health and 100 indicates the best possible health.[59] Lai and colleagues evaluated the association between the SF-36 and the SRS-22 and found that the SF-36 failed to capture important domains related to self-image.[60]

Due to limited published data, the reliability and validity of the SF-36 has yet to be established in the adult scoliosis population; however, it has been established in other populations. Test-retest

reliability of the SF-36 has been established for patients with Parkinson's disease, where for all domains of the SF-36, ICC ranged from 0.71 to 0.89.[61] In stroke patients, the SF-36 demonstrated adequate concurrent validity, as it correlated to the EuroQol instrument (r=0.66).[62]

EuroQol 5-Dimension Questionnaire (EQ-5D)

The EuroQol 5-Dimension Questionnaire (EQ-5D) is a widely used generic instrument measuring quality of life. The EQ-5D assesses five domains using a single question for each, these include: mobility, self-care, usual activities, pain/discomfort, and anxiety/depression.[63] The magnitude of each domain can be described by one of five possible levels: no problems, slight problems, moderate problems, severe problems, and extreme problems.[63] The scaling of the EQ-5D is scored from 0 to 1, where 0 indicates worst health and 1 indicates best health.[63]

No studies have assessed the validity and reliability of the EQ-5D in adults with scoliosis. However, in a study conducted by Cheung and colleagues, the Chinese version of the EQ-5D showed satisfactory validity and reliability in patients with adolescent idiopathic scoliosis (AIS).[64] Specifically, the authors found a significant correlation between domain scores of the EQ-5D (r = 0.57-0.74) and domain scores of the SRS-22r, which were intended to measure similar constructs.[64] Furthermore, the EQ-5D showed good test-retest reliability (ICC = 0.77).[64] In contrast, Adobor and colleagues[65] showed that the construct validity of the Norwegian version of the EQ-5D was poor to moderate when its domains were compared to the SRS-22 domains, with Pearson's r ranging from 0.14 to 0.58.

Numeric Pain Rating Scale (NPRS)

The Numeric Pain Rating Scale (NPRS) is a measure of pain intensity. It is an 11-point scale with individual question scores ranging from 0-10.[4] The NPRS is interpreted as: 0=no pain, 1-3=mild pain, 4-6=moderate pain, and 7-10=severe pain.[66] No studies have assessed the reliability and validity of the numeric pain rating scale for scoliosis.[5]

1.6.2 Specific Instruments

Specific questionnaires are instruments that are designed for specific diseases or groups of diseases.[57] They are based on the characteristics of the disease, and they attempt to highlight the effects caused by the disease or deformity.[56]

Oswestry Disability Index (ODI)

The Oswestry Disability Index (ODI) is one of the most widely-used tools for assessing lower back pain and disability.[14,67] The ODI is comprised of ten questions related to activities, performed over the preceding four weeks, that might have been affected by lower back pain.[68] Questions are scored from 0 to 5, and the composite sum score is doubled to provide a score of 0 to 100.[68] A higher score indicates worsening disability.[68] Various studies have shown excellent test-retest reliability of the ODI in patients with lower back pain (ICC estimated to be between 0.83-0.97).[69,70] However, the specific analysis on the use of the ODI on adults with scoliosis is limited.[54]

Scoliosis Research Society (SRS) Questionnaires

The Scoliosis Research Society (SRS) developed an instrument, the SRS-24, that was the first disease-specific PROM to evaluate changes to health-related, quality of life in patients with

adolescent idiopathic scoliosis who had undergone surgical intervention.[71,72] Some of the questions were derived from the SF-36.[73] The SRS-24 was revised to improve its psychometric properties, and became the SRS-23, which subsequently became the SRS-22.[74] However, the internal consistency of one of the domains of the SRS-22, namely the function domain, was found to be significantly low in individuals younger than 18.[57] Hence, the SRS-22 was modified to improve that domain, and the revised version is called the SRS-22r (refined).[75] The SRS-22r is a HRQOL questionnaire for adolescents with scoliosis.[71,75] The SRS-22r provides clinicians with a comprehensive evaluation of patient perceptions regarding the consequences of scoliosis and the effectiveness of treatments.[76] It consists of 22 questions across five domains: four five-question domains (measuring pain, self-image, function, and mental health) and one two-question domain (measuring satisfaction with management). The SRS-22r has been widely used to evaluate HRQOL in patients with adolescent idiopathic scoliosis. To calculate the Total score, the sum of the responses is divided by the number of questions answered. [51] Higher scores on all SRS questionnaires indicate better quality of life.[73,75]

Convergent validity has been evaluated for the SRS-22r in the adult scoliosis population. Both the self-image domain of the SRS-22r and the Total score of the SRS-22r correlated strongly with the Spinal Appearance Questionnaire v1.1 (r= -0.53 and r= -0.60, respectively), thus supporting convergent validity.[77] In contrast, the SRS-22 scores in the adult scoliosis population have demonstrated adequate discriminant validity by distinguishing between populations of affected and unaffected adults in the areas of function, pain, self-image, and mental health.[72] In addition, high test-retest reliability has been demonstrated in each domain of the SRS-22 (r= 0.83–0.94).[72]

However, in the study that assessed test-retest reliability of the SRS-22, a r-coefficient was used to measure reliability. This form of assessing reliability is not ideal because correlation coefficients only take into account the association between measurements without consideration to agreement between measures.[78] Furthermore, the mean Cobb angle measured for the participants was high (43.5 ± 20.9°), indicating limited representation of patients with mild curve severity.

The SRS-22 might not accurately assess health status in patients with milder forms of scoliosis, as the measure has a high ceiling effect (i.e. high proportion of participants with the best possible score).[79] In adolescents with idiopathic scoliosis, nine items of the SRS-22 have major (≥50%) ceiling effects, and 11 have moderate ceiling effects.[80] The high ceiling effects may negatively affect the ability of the SRS-22 to detect change when used in evaluative studies.

The Italian Spine Youth Quality of Life (ISYQOL) Questionnaire

The Italian Spine Youth Quality of Life (ISYQOL) questionnaire was recently developed to address the limitations of existing HRQOL tools that have been highlighted throughout this chapter.[81] In contrast to the SRS-22r, the ISYQOL was developed to measure changes related to varying levels of scoliosis treatment, ranging from conservative to surgical.[81] . Measurement scales developed with Rasch analysis have the attributes of a continuous measurement. Theoretically this provides more power when using it in statistical analyses.[81] The items of the ISYQOL were developed based on concerns expressed by the patients and from clinicians' input.[81] The items of the ISYQOL were developed based on concerns expressed by the patient and from clinician input.[81] To do this, content analysis was done for the concerns expressed by patients, parents of

patients, and scoliosis specialists in an online forum.[81] The result of the content analysis was a pool of 147 items. The appropriateness of these items was rated by clinicians and questions to address top ranked items were added to the ISYQOL. However, patients were not included in the rating process which potentially lead to the exclusion of items that may have been of higher importance to the patients.

The ISYQOL is a PROM that measures health-related quality of life in adolescents with idiopathic scoliosis.[81] The ISYQOL consists of 20 questions, 7 of which are answered only by brace wearers.[81] It provides a Total QOL score with lower scores representing higher quality of life.[81] The measurement properties of the ISYQOL have not been established in adults with scoliosis except for the internal consistency.[82]

Our lab has translated the ISYQOL to English from Italian using consensus provided by four team members. This translated ISYQOL was then reviewed by three Italian collaborators for compatibility with the original version.[83] The Italian collaborators found the English version to adequately represent the Italian version. In addition, our lab has tested the internal consistency of the translated ISYQOL in adolescents with idiopathic scoliosis and found that the translated ISYQOL met recommended standards for internal consistency (α= 0.79 - 0.84).[83] Zaina and colleagues evaluated the internal consistency of the same ISYQOL in adults with idiopathic scoliosis and also found it to be acceptable (α= >0.7). This suggested that the translated ISYQOL could be a useful tool to measure quality of life in adults with scolisis.[82]

1.6.3 Super-Specific Instruments

Super-specific questionnaires are instruments that have been developed to evaluate a specific dimension of scoliosis and to assess HRQOL in special populations of patients with scoliosis.[84] Instruments specifically evaluating body image represent a significant number of super-specific questionnaires used for scoliosis.[84]

Spinal Appearance Questionnaire (SAQ)

The Spinal Appearance Questionnaire (SAQ) is a tool that relies on pictograms to measure perceptions regarding the appearance of spinal deformity from patients and their parents.[85] The SAQ for patients consists of 20 questions (including 8 pictograms). These questions measure perceptions and expectations related to patient appearance, with higher scores indicating increasing negative perception regarding deformity.[85,86] Each item of the SAQ is scored from 1 to 5. Items are summed to produce 9 domain scores categorized as: General (items 9, 10, and 19), Curve (item 1), Prominence (items 2 and 3), Trunk Shift (items 4 and 5), Waist (items 11, 12, 13), Shoulders (items 6 and 16), Kyphosis (item 7), Chest (items 14 and 15) and Surgical Scar (item 17). Patients must also identify which two aspects of their deformity they find to be most bothersome (items 8 and 18). The last item is an open-ended question that asks the patient which aspects of their appearance they wish to change and asks for further elaboration[87].

A different version of the SAQ, namely the SAQ v1.1, presents a different scoring method that was introduced by Carreon and colleagues.[88] This new method calculates a Total score and only two domain scores: appearance (items 1-10) and expectations (items 12-15). No more than two

items can be unanswered for the appearance domain, and no more than one item can be unanswered for the expectation domain for scores to be considered valid. The scores are calculated as follows: the sum of relevant items available divided by five-times the number of items answered multiplied by twenty (to calculate the expectation score), fifty (to calculate the appearance score), or seventy (to calculate the Total score). The best possible Total score for the SAQ v1.1 is 14 and the worst possible score is 70.[88] A detailed comparison of the two versions of the SAQ will be presented in *Chapter 3*.

The SAQ provides more information regarding patient perceptions and concerns regarding scoliosis-related deformity compared to the SRS appearance domain.[89] The SAQ has a higher correlation coefficient to curve magnitude than the self-image domain of the SRS-22 for adolescents with idiopathic scoliosis (r= 0.36 compared to r= -0.20).[87] Additionally, in the adult population, the SAQ v1.1 correlated with both the self-image domain and Total score of the SRS-22r (r= -0.53 and r= -0.60, respectively), supporting convergent validity.[77] However, no published study has calculated the minimal detectable change of the SAQ.

One criticism of the SAQ is that it asks about patient perception of scoliosis-related appearance, as viewed from the back. This is not only impractical for patients, but this view may be less important to patients compared to their appearance as viewed from the front.[11] At the Edmonton Scoliosis Clinic, fewer patients complete the SAQ questionnaire than the SRS-22r questionnaire (per correspondence from Douglas Hill, a senior consultant at the clinic). Clinic staff report more patients have difficulty interpreting the pictograms used to assess appearance, and they find it easier to answer the non-pictogram questions from the SAQ or questions from the SRS-22r. This

information is consistent with findings reported by Mulcahey and colleagues.[90] In that study, adolescents with scoliosis were interviewed for their thoughts and opinions about the SAQ. Using this information, the researchers concluded that the SAQ uses complex medical jargon and asks vague questions that are difficult for patients to answer. These findings align with concerns expressed by the original authors of the questionnaire who pointed out as limitations, the potential for the SAQ to ask confusing and redundant questions.[85] Therefore, there is a need for a study to compare the two versions of the SAQ and assess whether the new SAQ features the same limitations of the old SAQ that had been expressed by patients.

The Body Image Disturbance Questionnaire-Scoliosis (BIDQ-S)

The Body Image Disturbance Questionnaire-Scoliosis version (BIDQ-S) is a self-reported, seven-item questionnaire that assesses patient concerns related to back shape and whether or not these concerns cause problems at school, work, or in interactions with friends and family. [91] Additionally, the BIDQ-S assesses whether patients avoid certain activities due to perceptions related to back shape. [91] The BIDQ-S is a modified version of the BIDQ. It includes 7 multiple-choice questions, where five response options focus on perceptions related to back appearance. The Total score is calculated by averaging the scores of questions 1A, 2A, 3, 4, 5A, 6A, and 7A.[91] Higher values indicate lower QOL. In adolescents with idiopathic scoliosis, the BIDQ-S showed internal consistency (Cronbach alpha =0.82).[91] Additionally, the BIDQ-S was significantly correlated with each domain of the SRS-22 and the SRS-22 Total score ($r = -0.50$ to -0.72, $p \leq 0.001$). Therefore, convergent validity exists between the BIDQ-S and the SRS-22 in adolescents

with idiopathic scoliosis.[91] Based on a current literature review, the BIDQ-S has not been validated for use in adults with scoliosis.

Truncal Anterior Asymmetry Scoliosis Questionnaire (TAASQ)

The Truncal Anterior Asymmetry Scoliosis Questionnaire (TAASQ) was recently developed to evaluate concerns of patients with adolescent idiopathic scoliosis. Questions are related to patient perceptions related to their anterior trunk appearance and the effects of these concerns on psychological mindset and behavior.[92] This tool includes 14 questions related to patient emotional status, specifically related to asymmetry of the breast, hip, and waist and to rib anterior prominence. The focus of the TAASQ on the anterior part of the body may enable it to address limitations of the SAQ.[92] Due to the novelty of the TAASQ, it has not yet been evaluated for use in the published literature.

1.7 **Problem Statement**

Documenting the measurement properties of PROMs is crucial to interpreting the outcomes of clinical treatments and research.[14] The choice of outcome measurement affects the validity of the inferences that can be drawn from any clinical research.[93] According to the Consensus-Based Standards for the Selection of Health Measurement INstruments (COSMIN), the quality of a measurement can be analyzed through assessment of measurement properties such as reliability

(consisting of reliability, internal consistency, and measurement error), validity (consisting of content validity, construct validity, and criterion validity), and responsiveness. [92]

Given the problems with existing scoliosis measurements and lack of evidence related to the metrology of the questionnaires reviewed above, there is need for studies to assess the measurement properties of newly developed PROMs. Through assessment, we can better ascertain if these new PROMs adequately assess scoliosis and if they address limitations featured in older PROMs. Because metrological properties may be sample dependent, it is important to compare multiple candidate PROMs in the same sample of adolescents and in the same sample of adults. Metrological evidence is also necessary to identify the best PROMs to use in the assessment of non-operative interventions for scoliosis because much prior evidence was obtained in candidates for surgery.

In this research study, the measurement properties from COSMIN will be used to compare new PROMs (ISYQOL, BIDQ-S, and TAASQ) to the widely used, scoliosis-specific PROMs (SRS-22r and SAQ v1.1) (**Appendix 1.2**). In addition, we will compare the new PROMs to radiographic measurements including the Cobb angle, vertebral rotation, and coronal balance. As previously established, we hypothesized that larger Cobb angles, a measure of scoliosis severity, will correlate with worsening quality of life score.[94,95] Furthermore, the new PROMs will be compared to the rotational deformity and to coronal balance, other important radiographic markers of scoliosis severity. It has been shown that vertebral rotation is associated with curve progression which might negatively affect patients' QOL.[27] Coronal balance might affect QOL in patients with scoliosis since it leads to contralateral knee and hip flexion.[96] To test for reliability, we will

measure the test-retest reliability of the new PROMs using two time-points. Finally, as recommended in the COSMIN, the minimum detectable change (MDC) will be calculated to provide initial information about the responsiveness of the new PROMs.

1.8 Thesis Flow

We found that despite the high prevalence of adult scoliosis, there is limited evidence reporting the effects of exercise on back pain, disability, and quality of life in adults with scoliosis. This suggests that further experimental research is needed. However, measures used to analyze the effectiveness of interventions in patients with scoliosis have important limitations or have not been validated for use in adults.

In this thesis, the *first chapter* is dedicated to providing background on the measures currently used to assess scoliosis, their limitations, and measurement properties assessing the metrology of these instruments. The *second chapter* of this thesis introduces a published systematic review which aims to review the effects of stabilization exercises on back pain, disability, and quality of life in adults with scoliosis.

Because this review found a paucity of evidence on the effect of exercises in adults, together with the limited metrology research summarised in chapter, the hypothesis that there is a lack of adequate determination of the measurement properties of tools to measure this population was formulated. The *third chapter* of this thesis seeks to compare the SAQ with a newly revised version, the SAQ v1.1 to determine which of these established questionnaires can be used as a reference tool in validity testing in the subsequent chapters. The next two chapters aim to fill the

gap in metrological evidence about new tools to assess quality of life and perceived appearance in adolescents and adults with scoliosis.

The *fourth chapter* of this thesis presents a study that evaluates the test-retest reliability and the convergent validity of the Italian Spinal Youth Quality of Life questionnaire (ISYQOL), Truncal Anterior Asymmetry Scoliosis Questionnaire (TAASQ), and the Body Image Disturbance Questionnaire-Scoliosis (BIDQ-S) questionnaire compared to the Scoliosis Research Society-22 (SRS-22r) and Spinal Appearance Questionnaires v1.1 (SAQ v1.1) for adolescents with scoliosis. Furthermore, this study compared the new PROMS to common radiographic measurements used in scoliosis research, including the Cobb angle, vertebral rotation, and coronal balance.

In the *fifth chapter*, this thesis assessed the measurement properties of the aforementioned questionnaires in adults with scoliosis. The goal of this section is to determine whether these PROMs are appropriate for use in adults with scoliosis since outcome measurements of QOL for this population are currently lacking.

Finally, the *sixth chapter* will summarize the results of the included studies, list the overall limitations, and suggest directions for future studies. Also, this chapter will discuss the results of the studies in light of the current literature.

2 CHAPTER 2 – EFFECT OF STABILIZATION EXERCISES ON BACK PAIN, DISABILITY, AND QUALITY OF LIFE IN ADULTS WITH SCOLIOSIS: A SYSTEMATIC REVIEW

This chapter has been published in the European Journal

of Physical and Rehabilitation Medicine (Alanazi et al., 2018)

Alanazi MH, Parent EC, Dennett E. Effect of stabilization exercise on back pain, disability and quality of life in adults with scoliosis: a systematic review. *Eur J Phys Rehabil Med*. 2018;54(5):647-653. doi:10.23736/S1973-9087.17.05062

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ABSTRACT

***Introduction*:** Adult Scoliosis (AS) is the most common spine deformity in adults. Back pain is the main symptom leading patients to seek medical consultation. Stabilization exercise has been shown effective for reducing back pain. No literature review has examined the effects of such exercises in adults with scoliosis. The objective was to systematically review the effects of stabilization exercises on back pain, disability, and quality of life in adults with scoliosis.

Evidence Acquisition: We systematically searched the following databases from inception to March 2017: Medline, CINAHL, Embase, SportDiscus and Cochrane Register of Controlled Trials. Selection criteria included: controlled clinical trials that compare core stabilization exercise to placebo, no treatment or another treatment in participants diagnosed with AS over 18 years old. Studies with participants presenting torso or lower extremity surgery, injection in the last six months, comorbidity that could affect the spine, red flags signs or with a history of spine trauma were excluded. We extracted information about participants, treatments, and results on pain, function and quality of life. We appraised quality using Cochrane risk of bias. We formulated level of evidence summary using a priori rules based on quality and consistency of results.

Evidence Synthesis: We found 630 unique articles and screened the full-text of 98 articles retrieved. Only one article met all selection criteria and was included in this review. Studies were mainly excluded for study design and patient population. The included study presented a low risk of bias for all criteria except for blinding and reporting if the timing of assessments was similar between groups. The literature provides limited evidence from one study with high risk of bias that

stabilization exercises in the form of 20 weeks of active self-correction, task-oriented exercises and cognitive–behavioral therapy significantly improves pain, disability and quality of life.

Conclusion: Despite the high prevalence of AS, there is an important gap in the literature with limited evidence reporting the effect of exercise on back pain in adult with scoliosis. This review suggest further experimental research is needed and formulates research recommendations.

2.1 Introduction

Scoliosis, the most common spinal deformity affecting adults, is defined as a complex deformity of the spine that develops in 3-dimensions and leads to frontal curves, vertebral rotations, and a flattening of the sagittal physiological curves.[97] When scoliosis develops after growth is completed (after 18 years of age), it is called Adult Scoliosis (AS). AS is defined as a spinal deformity in a skeletally mature patient with a Cobb angle of more than 10 degrees in the coronal plane[98].

AS is getting more attention mainly due to the demographic shift toward an older population, the patients' awareness of natural history, and their willingness to overcome chronic pain and limitations in activities[99]. The prevalence of AS ranges from 1.4 to 20 % in the adult population and AS affects as many as 68% of the individuals over 60 years of age[99,100].

Patients with AS present with various symptoms, including pain, curve progression, symptomatic radiculopathy, or cosmetic deformity affecting the quality of life and physical function[101–103]. However, pain is the most common symptom and occurs in approximately 90% of patients with AS[102]. Low back pain is prominent in patients with AS, resulting from muscle fatigue and muscle spasm over the convexity of the curvature[28].

Therapeutic options to manage AS are either operative or non-operative. Although operative options have been suggested to be the superior intervention, physicians use caution when considering surgery because of the associated risk of complications in the adult population[101,104]. Further, to our knowledge, to date there are no head to head comparisons of operative and non-operative treatments in this population and only a few high-quality studies of non-operative

interventions. Therefore, understanding the effects of non-surgical treatments options such as physical therapy, chiropractic or manipulation, and injection or epidural for AS is required[101].

Exercise regimens that focus on core strengthening or core stability are among recommended interventions in AS management[105]. Core exercise is defined as “the restoration or augmentation of the ability of the neuromuscular system to control and protect the spine from injury or reinjury” and "used to describe a spectrum of exercise approaches that have the common goal to improve lumbopelvic control with varied rationales[44].” A systematic review has focused on treatment options for AS[106] but did not specifically address the effect of stabilization exercise on back pain in AS. Core stabilization exercise have been shown effective for improving pain and function in patients chronic low back pain [28,46,47,48], however, the literature comprises unclear evidence to support the effects of these exercises in the population of adults with scoliosis. Therefore, this study aims to systematically review the published evidence to determine if core stabilization exercise is a viable alternative treatment for adults with scoliosis.

The primary aim of this systematic review is to evaluate the effect of the stabilization exercise on back pain in adult with scoliosis. The secondary aim is to evaluate the effect of stabilization exercise on the quality of life and disability levels in adults with scoliosis.

2.2 Evidence Acquisition

The protocol for this systematic review is available in the PROSPERO database (CRD42017060805).

We conducted a systematic search of the following databases: Medline (OVID), CINAHL (EBSCO), Embase (OVID), SportDiscus (EBSCO) and the Cochrane Central Register of Controlled Trials (CENTRAL). The search was carried out from inception to March 9, 2017 in each database. The search strategy was developed using the PICOS framework by the authors, who are expert clinicians and researchers in the field of scoliosis. A librarian (LD) identified corresponding indexed terms and carried out the search within each of the databases. The search strategy included a combination of subject headings and keywords combining the concepts of scoliosis to define the population and stabilization exercise as the treatment of interest. Terms related to outcomes and comparison interventions were not specified in the search to ensure we captured all relevant research using the outcomes of interest and all compared interventions. The search was limited to the English language and we excluded conference abstracts and letters. The full search strategy used for each database is available in Appendix (1).

2.2.1 Study Selection Criteria

Types of Studies

We included randomized controlled trials (RCTs), prospective controlled clinical trials, and retrospective controlled studies because it was anticipated that very few RCTs would be identified. Studies with less than 10 participants per group were not included.

Types of Participants

Participants had to be diagnosed with AS, have a Cobb angle of 10º or more, and be 18 years of age or more. Because the majority of the studies do not provide details of de novo/ degenerative scoliosis (occurs through degenerative change without preexisting spinal deformity)[102] or idiopathic scoliosis (a continuation of adolescent idiopathic scoliosis)[107] presentation, both types of scoliosis were included. In addition, we only included studies with participants that did not have exercise treatment in the three years before the study and that examined the effect of the stabilization exercise therapy meeting the definition of core exercises presented in the introduction. In studies with a mixed age group, at least 75% of the sample had to be over 18 years of age and have a diagnosis of AS.

Studies with participants presenting any torso or lower extremity surgery or any injection in the last six months were excluded. Similarly, participants with any comorbidity that could affect the spine, red flags signs or with a history of spine trauma were excluded[108].

2.3 Types of Interventions

2.3.1 Experimental Intervention

The experimental intervention of interest in this review included all types of exercises meeting the definition of core exercises stated in the introduction[44]. Exercise that was combined with other types of intervention were also included.

2.3.2 Comparison Interventions

Comparison interventions were not restricted and could include: placebo, no treatment, drug management, spinal injection, spine surgery, or any other type of nonsurgical treatments (e.g. braces, electrical stimulation, chiropractic, manual therapy, manipulation and mobilization).

2.4 Types of Outcome Measures

This review mainly assesses the effect of the stabilization exercise on back pain. Back pain, the primary outcome, could be measured using numerical rating scale (NRS)[109] or other validated measurement tools, such as pain drawing, or pain pressure threshold.

Quality of life and disability, the secondary outcomes, were included as measured by specific validated questionnaires, such as Oswestry,[110] Roland – Morris,[111] Scoliosis Research Society Outcomes Questionnaire (SRS-22),[112] 36-Item Short Form Health Survey (SF-36),[113] Brace Questionnaire (BrQ),[114] and Scoliosis or the Quality of Life Index (SQLI)[115,116].

2.5 Data Collection and Analysis

2.5.1 Selection of Studies

Search results were uploaded into RefWorks reference management software (RefWorks version 2.0, ProQuest, Ann Arbor, Michigan, United States) and duplicates were removed. Screening was completed in two stages using Covidence [117]. Titles and abstracts were screened for eligibility by two reviewers assessing all references. The reviewers were two Physical therapists with 5 to 21

years of clinical and research experience. Full texts of potentially relevant studies were obtained, uploaded into covidence, and assessed independently for inclusion by two reviewers. Reasons for exclusion were tracked at the full-text stage. A third reviewer would have been contacted to resolve disagreement if consensus discussion between reviewers did not resolve disagreements (but was not ultimately needed).

2.6 Data Extraction

Two researchers (MA and EP) independently extracted results from the included studies using the modifiable extraction form in Covidence. Extracted data included study design, patient characteristics (number of participants, age, sex, Cobb angle, BMI, weight, height), description of the experimental and comparison interventions, co-interventions, adverse effects, duration of follow-up, outcomes assessed and results.

2.7 Risk of Bias Assessment

Two review authors (MA and EP) assessed the risk of bias for the included study by utilizing the Cochrane risk of bias assessment tool which was available through Covidence. We added the following criteria to the Cochrane risk of bias assessment tool based on recommendations from the Cochrane back group: timing of outcome assessment similar, similarity of baseline prognostic indicators, co-intervention avoided or similar, and compliance acceptable[118]. Conflicts were solved by the senior reviewer (EP). The risk of bias assessment included 13 relevant criteria (**Table 2-1).** Each criterion was scored as presenting high, low, or unclear risk of bias. A study was judged to be of high quality if all the criteria were reported to have low risk of bias[119].

2.8 Data Synthesis

Summary statements were formulated combining quality appraisal information with details about the consistency of the results as per the following rules. **Strong** evidence corresponds to consistent results (75% or more) from at least 2 high quality studies. **Moderate** evidence corresponds to consistent results (75% or more) in 1 or more low quality studies and 1 high quality study. **Limited** evidence corresponds to findings from 1 high quality study or consistent results (75% or more) among low quality studies. No evidence is used when no study is identified. A **Conflicting** level of evidence corresponds to inconsistent results (>25%) irrespective of study quality[120]. A meta-analysis was not planned for this review as the level of heterogeneity in population, settings and intervention used was anticipated to be high.

2.9 Evidence Synthesis

Using our search strategy, we found 908 articles, resulting in 630 articles after excluding duplicates. After screening the titles and abstracts, only 105 articles were included for full-text screening. A Total of 98 full-text articles could be retrieved and after screening, only one article fit the selection criteria and was included. The main reasons for exclusion were study design and patient population. (PRISMA flow chart **Figure 1).**

The Risk of Bias assessment of the included study is reported in **Table 2-1**. The included study presented overall a high risk of bias because of the lack of blinding. Authors also did not report if the timing of assessments was similar between groups. For all other criteria, the study presented a low risk of bias.

2.10 Included Study

Monticone et al. completed a randomized controlled trial with a sample of 130 adults with scoliosis[104]. Patients who underwent a 20-week rehabilitation program consisting of (1) active self-correction, task-oriented exercises and (2) cognitive–behavioral therapy were compared to a control group treated with "general physiotherapy [that] included exercises for spinal mobilization (passive mobilization to improve thoracic and lumbar range of motion), muscle segmentary stretching of upper/lower limb and back muscles, strengthening of abdominal and back muscles, and postural control (involving exercises aimed at developing motor control of the spine and pelvis)"[104]. Patients in each group had one 60-min session of physical training per week for 20 weeks. In addition, the experimental group met with the psychologist twice a month for a 60-minute session. The participants' characteristics were similar between groups (**Table 2-2)**. These participants presented a moderate baseline level of disability and pain.

2.11 Level of Evidence

The extracted results for the outcomes of interest from the included study are presented in **Table 2-3**. The literature provided limited evidence from one study with high risk of bias that stabilization exercises in the form of 20 weeks of active self-correction, task-oriented exercises and cognitive–behavioral therapy significantly improved pain measured using the numeric pain rating scale more than general physiotherapy (difference of 3.2/10 between group after the program). There was also a limited level of evidence demonstrating significant improvements following 20 weeks of active self-correction, task-oriented exercises in quality of life measured using each domain of the SRS -

22 questionnaire (differences between groups after the program: 0.7/5 for function, 0.9 for pain, 0.5 for self-image, 0.7 for mental health, and 1.0 for satisfaction with care) and in disability levels measured using the Oswestry questionnaire (12% difference between groups after the program).

2.12 Discussion

Back pain is the main driving symptom that leads patients to seek medical consultation in adult scoliosis[102]. Because of the high risk of complications associated with surgery and population aging, non-operative interventions are more utilized in treating adult with scoliosis[101,104].

Only 1 study[104] was ultimately included in this review documenting the effect of a form of stabilization exercises on outcomes of interest in AS. However, there are studies in adolescents with idiopathic scoliosis[42,43,121–125] that, consistent with the findings of the included study, also show beneficial effects of exercises which were not included in this review because the population is different from our interest. Some case studies also support the effect of stabilization exercises in adults with scoliosis but this study design was not included in our review or these reports did not measure the outcomes of interest and instead reported positive effects on outcomes such as Cobb angle[126] and chest expansion[127]. Furthermore, we found a review of studies that investigated the effect of physical exercise but did not specify what type of exercise was reviewed[101].

Operative options have been suggested to be superior to non-operative care [101]. Yet, we could not find any prospective head-to-head studies comparing the different types of non-operative interventions (especially exercise) to surgery to support this superiority claim. Since failure of appropriate conservative options is often a prerequisite to surgical intervention in many centers,

such comparisons may be quite difficult to perform[101]. While some retrospective studies reported positive outcomes of surgical care relative to non-operative care, the non-operative care was often not clearly defined[128]. Glassman et al. documented non-surgical resource utilization in groups of adults with spinal deformity with high and low symptoms and found that only 38% and 33% of the participants used exercise in each group, respectively. Non-operative care also included a wide range of 16 other treatments than stabilization exercises and dosage was not documented[129].

Scheer et al. and Bridwell et al. also compared outcomes in patients with adults spinal deformity receiving surgery or non-operative care carefully conducting propensity matching and found results favoring surgery.[128,130] However, in both studies, the recruitment of the non-operative cohort occurred at the surgery clinic among patients being seen for a possible surgical intervention. The non-operative care was also not homogenous among participants and prescribed individually. Most importantly though, Scheer et al. did not define non-operative care at all and did not report the number of patients receiving different types of treatments. Bridwell et al. reported that the non-operative treatment included observation (21%), medications (26%), medications plus (physical therapy and/or injections) (40%), and other treatment without medications (13%). It is obvious that the majority of patients did not receive exercise treatment. In both studies the dosage of each non-operative treatment was also not monitored. These examples and the gap found in the present review illustrate the need to investigate the effectiveness and cost efficiency of different types of clearly defined non-operative interventions including core stabilization exercise to help patients, physicians and physical therapist choose appropriately.

Despite finding one RCT, we only have a limited level of evidence that stabilization exercise has positive effect on pain, disability and all domains of the SRS-22 quality of life questionnaire. This conclusion is based on one study that has a low quality because it has a high risk of bias because of lack of blinding of the participants and assessors. However, blinding is known to be hard to apply in physical therapy studies. Otherwise this RCT presented low risk of bias in supporting positive effects of exercises on the outcomes of interest.

Monticone et al.'s result demonstrated good outcomes in specialized centers and by physiotherapists offering a stabilization exercise program tailored specifically to scoliosis which is different from what the majority physiotherapists are trained to offer for patients with low back pain. Therefore, the generalizability of this approach may be limited to specialized centers and physiotherapists specifically trained in delivering scoliosis-specific exercises.

In the future, studies are needed that clearly create a sample that does not mix participants diagnosed with AIS and with degenerative scoliosis. This would help drawing more accurate conclusions in the event where these two groups would present a different prognosis. Also, the Consensus between SOSORT and SRS non–operative management committee[131] highly recommends that "prognostic factors for consequences of the deformity in adulthood on primary patient-centred outcomes (such as aesthetics, deformity progression, disability, pain and quality of life) be continuously researched and better defined by high quality studies"[131]. They also recommend subgroup reporting based on curve severity for radiographic research outcomes. In regard to when the outcomes should be reported, it would be important if future studies have roughly similar duration so that they can be comparable.

2.12.1 Limitations of the Study

Some methodological limitations may affect the interpretation of the results from this review. Relevant articles may have been missed because only articles written in English in databases capturing mostly literature in English were included. The small number of studies ultimately found is a limitation as it prevented reaching higher levels of evidence, examining reviewer agreement on selection and quality appraisal, as well as, prevented attempting a meta-analysis.

2.13 Conclusion

Stabilization exercise, as reported in the included study, is shown to be effective in reducing back pain, disability and improving quality of life in adults with idiopathic scoliosis. However, this review highlights the paucity of literature examining the effect of exercise on back pain in adult with scoliosis and strongly suggests that further experimental research is needed aiming to ensure proper blinding as this was a common weakness.

2.14 Funding

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2.15 Acknowledgements

The authors would like to express their thanks Dr. Sanja Schreiber for her help with Covidence and the search.

Table 2-1: Risk of Bias Assessment

CRITERIA	SCORE
Sequence generation	LOW
Allocation concealment	LOW
Blinding of participants and personnel	HIGH
Blinding of outcome assessors	HIGH
Incomplete outcome data	LOW
Selective outcome reporting	LOW
Timing outcome assessments similar?	UNCLEAR
Similarity at baseline characteristics?	LOW
Co-intervention avoided or similar?	LOW
Compliance acceptable?	LOW
Blinding of care provider to the intervention?	HIGH
Randomized participant analyzed in the group to which they were allocated?	LOW
Other sources of potential bias?	LOW

Table 2-2: Baseline Characteristics of the Participants in the Included Study

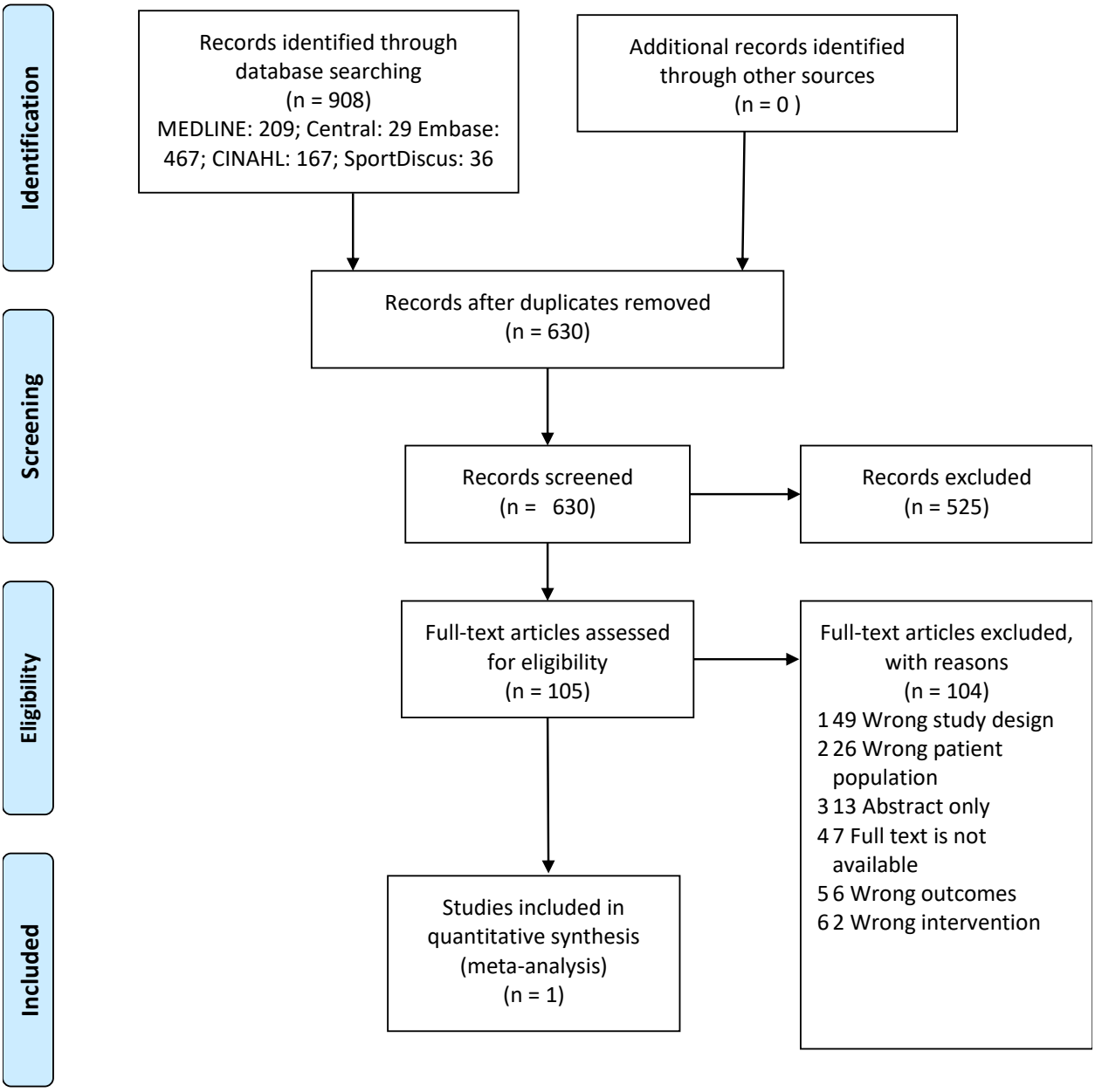
Characteristic	Experimental group	Control group
Age (years)*	51.6 (8.1)	51.7 (8.5)
Gender (male/female) *	17/48	19/46
Body mass index (kg/m2) *	21.8 (3.7)	22.0 (3.5)
Pain duration (months) *	37.9 (20.5)	35.4 (19.9)
Pain radiation to lower limbs (yes/no)	25/40	30/35
Type of scoliosis (Thoracic)	15	14
Type of scoliosis (Lumbar)	20	19
Type of scoliosis (Thoracolumbar)	30	32
Main curve (Cobb angle)	28.2 (4.9)	27.5 (5.0)

*Mean values (standard deviation)

Table 2-3: Extracted Results from the Included Study

Outcome	**Group**	**Baseline** Mean value (standard deviation	**Pre-training** Mean values (standard deviation)	**Post-training** Mean values (standard deviation)
Numeric Pain rating scale **(0-10)**	Experimental (n=65)	6.5 (1.2)	2.9 (1.2)	2.2 (1.2)
	Control (n=65)	6.6 (1.2)	6.1 (1.4)	6.3 (1.7)
Oswestry disability index **(0–100)**	Experimental (n=65)	38.0 (6.8)	19.7 (6.4)	17.6 (5.3)
	Control (n=65)	37.8 (6.0)	31.8 (6.1)	32.5 (6.5)
SRS-22 **(Function 0-5)**	Experimental (n=65)	2.7 (0.5)	3.7 (0.5)	4.2 (0.3)
	Control (n=65)	2.7 (0.5)	2.9 (0.3)	2.9 (0.4)
SRS-22 **(perceived Self-image 0-5)**	Experimental (n=65)	2.9 (0.4)	3.5 (0.6)	3.8 (0.5)
	Control (n=65)	2.9 (0.4)	2.9 (0.4)	2.8 (0.4)
SRS-22 **(Mental health 0-5)**	Experimental (n=65)	3.4 (0.4)	4.2 (0.4)	4.4 (0.3)
	Control (n=65)	3.5 (0.4)	3.5 (0.4)	3.5 (0.4)
SRS-22 **(Pain 0-5)**	Experimental (n=65)	2.8 (0.5)	3.8 (0.5)	4.2 (0.4)
	Control (n=65)	2.8 (0.5)	2.9 (0.4)	2.7 (0.5)
SRS-22 **(Satisfaction with management 0-5)**	Experimental (n=65)	NA	4.5 (0.2)	4.5 (0.2)
	Control (n=65)	NA	3.5 (0.3)	3.0 (0.5)

Figure 2-1: PRISMA Flow Chart



3 CHAPTER 3 – TEST-RETEST RELIABILITY AND CONVERGENT VALIDITY OF THE SPINAL APPEARANCE QUESTIONNAIRE IN ADOLESCENTS WITH IDIOPATHIC SCOLIOSIS

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Abstract

Introduction: Patients with adolescent idiopathic scoliosis (AIS) risk developing symptoms in their adult lives related to decreased quality of life, cosmetic deformity, pain, and progressive functional limitations. The Spinal Appearance Questionnaire (SAQ) was developed because the patients' perceptions of their appearance influenced by scoliosis are difficult to assess. The SAQ has been modified to a newer version, the SAQ v1.1, in order to address previous limitations such as unclear scoring strategy and ceiling effect. There is need to compare the measurement properties of the SAQ and the SAQ v1.1.

Objective: This study aims to compare the test-retest reliability and convergent validity of the two scoring versions of the SAQ in patients with AIS. Our secondary objective was to determine the ceiling and floor effects of both versions.

Study Design*:* A metrological study was conducted using a cross-sectional design for the assessment validity and using a single-group, two repeated-measures design for reliability.

Sample*:* One-hundred females with AIS (n = 100), aged 10-18 years old, treated non-operatively for scoliosis, were recruited from a Canadian pediatric scoliosis clinic.

Outcome Measures: Three scoliosis specific questionnaires (Scoliosis Research Society-22 refined (SRS-22r), Spinal Appearance Questionnaire (SAQ), and Spinal Appearance Questionnaire v1.1 (SAQ v1.1)) were collected along with radiographic measurements (maximum Cobb angle, coronal balance, as well as thoracic and lumbar vertebral rotations).

***Methods**:* Questionnaires were administered twice electronically with a one-week interval between.

Results: Participants included 100 females with a mean age of 13.9±1.8 years and a curve angle of 28.8^{o} ±13.9^{o}. Intra-class correlation coefficients ($ICC_{(3,1)}$) varied between 0.72 to 0.94 for the test-retest reliability of the SAQ domains. The ICCs for the scores of the newer SAQ v1.1 (Total, Appearance, and Expectations domains) were: ICCs (95%CI) = 0.92 (0.87-0.96), 0.94 (0.89-0.97), and 0.86 (0.75-0.92), respectively. Convergence with SRS-22r Total score and Cobb angle was supported for the SAQ (r = 0.32 to 0.59). Convergence for the SAQ v1.1 Total score with the SRS-22r Total score was shown with a Pearson r = -0.50. Convergence for the SAQ v1.1 and the Cobb angle was r = 0.63. Ceiling and floor effects were observed only for the following domains of SAQ domains: Curve= 11% (ceiling effect), Kyphosis = 68% (ceiling effect), and Waist = 4% (floor effect). Ceiling and floor effects for the SAQ v1.1 domains were: Total = 3% (ceiling effect), Appearance = 5% (ceiling effect), Expectations = 14% (ceiling effect), and Expectations = 10% (floor effect).

Conclusion: We support the use of the SAQ v1.1 over the SAQ in both research and clinical practice as the SAQ v1.1 has stronger reliability, stronger convergent validity, and fewer ceiling and floor effects in patients with AIS. Future studies should further investigate the responsiveness of the SAQ v1.1.

3.1 Introduction

In patients with adolescent idiopathic scoliosis (AIS), there is a higher risk of developing scoliosis-related health problems such as having decreased quality of life, cosmetic deformity, pain, and progressive functional limitations in adulthood, especially if the curve magnitude is 30° or more at skeletal maturity.[26] If the curve is over 50°, it is almost certain that the curve will progress throughout adulthood, leading to deterioration of health-related quality of life. [26]

*Health-related quality of life (*HRQOL) is an assessment of the effects of a disease or treatment on the physical, psychological, and social domains of functioning and well-being of a patient.[55] HRQOL in scoliosis is influenced primarily by disease-related changes to appearance.[55] Appearance is one of the main reasons that patients with AIS undergo surgery.[5] Additionally, patients with AIS may suffer from psychological symptoms related to depression and low self-esteem due to scoliosis-related changes to their appearance. [5,39] Current instruments used for patients with AIS such as the Spinal Appearance Questionnaire (SAQ) show limitations in their measurements properties.

The SAQ assesses patient perception of their own appearance as viewed from the back.[85] The SAQ was created as a modification of the Walter Reed Visual Assessment Scale (WRVAS). The WRVAS is a seven-item questionnaire where each item can be answered by choosing one of five standardized figures representing varying degrees of deformity.[86] The original WRVAS items were: body curve, rib prominence, flank prominence, head rib pelvis, head pelvis, shoulder level, and scapula rotation. Sanders and colleagues[85] modified the WRVAS to develop the SAQ. The

SAQ consists of 20 items that are summed to produce nine domain scores: General, Curve, Prominence, Trunk Shift, Waist, Shoulders, Kyphosis, Chest and Surgical Scar. Patients must also identify two aspects of their deformity that they find most bothersome. The last item on the SAQ is an open-ended question that asks patients which aspects of their appearance do they most wish to change and why.[11] However, through interviewing adolescents with scoliosis, Mulcahey and colleagues[90] found that most patients found the SAQ to be overly complicated by the use of jargon and confusing due to the vagueness of the questions. For example, participants found it difficult to differentiate between breast and chest, and hips and waist. Overall, they do not support the use of the SAQ for evaluating changes related to scoliosis. These findings are consistent with concerns expressed by the original creators of the SAQ, who described the potential for confusion and redundancy in their published findings.[85]

Most recently, Carreon and colleagues [88] developed a revised version of the SAQ named the Spinal Appearance Questionnaire version 1.1 (SAQ v1.1) to improve items distribution and scoring strategy . Using factor analysis, they modified the questionnaire to contain 14 items loaded on two factors. The summation of the first 10 items are related to the Appearance domain and the summation of the last 4 items gives the Expectations domain. Contrasting with the old SAQ, the SAQ v1.1 can generate a total score through the addition of both domains. This total score can range between 14 and 70, where the lower scores represent positive perceptions of self-image.

Given the limitations of the SAQ, there is a need to study and compare the measurement properties of the SAQ and the SAQ v1.1. This is necessary to determine whether or not the SAQ v1.1 adequately resolves the limitations of the old SAQ. Additionally, the SAQ v1.1 can be assessed

to see if it meets minimum standards for measurement properties the SAQ v1.1 can be assessed to see if it meets minimum standards for measurement properties proposed in the Consensus-Based Standards for the Selection of Health Status Measurement Instrument (COSMIN) for use in research and clinical settings.

3.2 Objective

This study aims to compare the test-retest reliability and convergent validity for the SAQ and SAQ v1.1 in patients with AIS. Additionally, this study seeks to determine the ceiling and floor effects for both as a secondary objective.

3.3 Study Design

A metrological study was conducted on a single group using a cross-sectional design to test for validity and ceiling and floor effects. Data was collected once and then repeated once one week later to test for reliability.

3.4 Hypothesis

We hypothesized that:

1. The test-retest reliability of the SAQ v1.1 scores will be adequate and meet the minimum acceptable standards suggested by COSMIN for use in groups of patients ($ICC_{3,1} \geq 0.7$) and individual patients ($ICC_{3,1} \geq 0.90$).[132]

2. The SAQ v1.1 score indicating better QOL will have a correlation coefficient ≥ -0.35[133] with the SRS-22r. Additionally, the SAQ v1.1 will have a higher correlation with the SRS-22r self-image domain, which will be equally or better correlated with radiographic measurements.

3. The scores of the SAQ v1.1 will be free from ceiling and floor effects or have less ceiling or floor effects than the SAQ (less than 15%).[132]

These hypotheses are based on the assumption that larger values for radiographic measurements should be correlated to poorer quality of life and more negative perception of self-image.[94,95]

3.5 Sample

Eligible participants were recruited for this study from the Stollery Children's Hospital while attending routine scoliosis clinic visits. Informed consent was obtained from eligible participants and their legal guardians. This study, including its consent documentation, was reviewed and approved by the University of Alberta Health Research Ethics Board (Pro00073569). A total of one-hundred participants (n=100) were recruited for this study. *Inclusion criteria* for this study included: having an active diagnosis of idiopathic scoliosis, be of 10-18 years of age, have curve severity over 10°, and being fluent in English. *Exclusion criteria* included: having a history of spine surgery, having a history or active diagnosis of other diseases that might affect quality of life and/or torso appearance, or having history of trauma in the torso or lower extremities.

3.6 Data Collection Procedures

Questionnaires were programmed in the Research Electronic Data Capture (REDCap) software and administered as an online survey (version 7.6.9). Using the automatic invitation feature in REDCap, participants completed questionnaires online after one week of the clinic visit and after two weeks. Participants required roughly 15 minutes to complete all five questionnaires each time. To minimize missing data, an automatic prompt appeared if any questions are skipped during response submission.

At the start of the study, standing posterior-anterior radiographs of the full spine were obtained using the EOS system. The Cobb angle, axial vertebral rotation, and coronal balance were subsequently measured by one rater (MA) using custom Medical Image Analysis Software (MIAS) software (version 9.6.7.0). Prior to data collection, MA, a physical therapist, was trained to collect this data in a uniform manner by observing one demonstration, discussing ten practice cases, and conducting these measurements twice ($ICC_{3,1}$= 0.99, 95% CI .94 to 1).

Radiographic files were uploaded to MIAS after being converted from Digital Imaging and Communications in Medicine (DICOM) to Joint Photographic Experts Group (JPEG) image format using ImageJ software (version 1.50i) (Rasband, W.S., ImageJ, U. S. National Institutes of Health, Bethesda, Maryland, USA, https://imagej.nih.gov/ij/, 1997-2018). The evaluator of radiographic measurements was blinded to the identity of the participants and to questionnaire scores during image measurement.

3.7 Outcome Measures

3.7.1 Spinal Appearance Questionnaire (SAQ)

The Spinal Appearance Questionnaire (SAQ) is a pictorial scale that measures patients perception regarding their appearance as it relates to spinal deformity.[85] The SAQ consists of 20 items, eight of which are pictograms, that describe various components of spinal deformity.[85] Five response options are provided as drawings that have depictions ranging from mild to severe deformity (scores 1 to 5). The remaining questions can be answered by choosing one of the following options: 'Not true' (1), 'A little true' (2), 'Somewhat true' (3), 'Fairly true' (4), and 'Very true' (5).[85] Answers for each domain are summed to provide a total score for each domain, ranging from 1 to 5, where 1 indicates good QOL.[85] This version of the SAQ does not feature a total score.

3.7.2 Spinal Appearance Questionnaire Version 1.1 (SAQ v1.1)

Spinal Appearance Questionnaire version 1.1 (SAQ v1.1) is a modified version of the SAQ. It consists of 14 items. The first ten items are related to the 'Appearance' domain and the last four items are related to the 'Expectations' domain. The ten items related to the appearance domain are as follows: body curve, rib prominence, flank prominence, head-chest-hips, position of head over hips, shoulder level, shoulder blade rotation, shoulder angle, head position, and spine prominence. The four items related to the expectations domain are as follows: 'I want to be more even'; 'I want to have more even shoulders'; 'I want to have more even hips', and 'I want to have a more even waist'. For the 'Appearance' domain, response options are provided as drawings ranging from mild (score of 1) to severe (score of 5). The 'Expectations' domain features verbal response options ranging from 'not true' (score of 1) to 'very true' (score of 5). Answers are subsequently summed

to provide a score ranging from 10 to 50 for the 'Appearance' domain and ranging from 4 to 20 for the 'Expectations' domain. The SAQ v1.1 Total score is calculated by summing the total scores of both domains.[88] The best possible total score for the SAQ v1.1 is 14, related to better QOL and more positive perception of scoliosis related deformity, and the worst possible score is 70.[88] In a study conducted by Carreon and colleagues, 1,802 adolescents with AIS were given the SAQ v1.1 and their results suggested that the SAQ v1.1 had good reliability (Cronbach's $\alpha \geq 0.88$; test-retest correlation ≥ 0.81) and convergent validity as it relates to major curve magnitude ($r=0.32 – 0.36$).[88]

3.7.3 Scoliosis Research Society-22 Refined Questionnaire (SRS-22r)

The Scoliosis Research Society-22 refined questionnaire (SRS-22r) is the most commonly used instrument for measuring HRQOL in patients with scoliosis.[75] It consists of 22 questions covering five domains: Pain, Self-Image, Function, Mental Health, and Satisfaction with Management. Scores range from 1 to 5 per question, and these scores are averaged to produce domain scores and a total score.[75] Higher scores on the SRS-22r indicate better quality of life. The SRS-22 demonstrated acceptable reliability ($ICC_{3.1}$ = 0.85 to 0.96) and concurrent validity ($r > 0.70$) compared to the Short Form 36 (SF-36). However, 56.9% of respondents were given the highest score possible, showing that this assessment has ceiling effect for patients with post-operative idiopathic scoliosis.[75] The SRS-22 has been refined to improve the internal consistency of its Function domain, increasing to $\alpha=0.67$ to 0.78 following changes.[134]

3.7.4 Cobb Angle

To obtain the Cobb angle for each patient, a straight line was drawn along the upper endplate of the most tilted vertebra above the apex and along the lower endplate of the most tilted vertebra below the apex for each curve. The apex was identified by choosing the most translated vertebra in each of the spine curvatures. The angle between these two lines forms the Cobb angle.[24] For our participants, all curves were measured and entered in the data file along with location of the selected endplates and apex of each curve.

3.7.5 Axial Vertebral Rotation (AVR)

Axial vertebral rotation is the rotation of a vertebra around its longitudinal axis when projected onto the transverse image plane.[135] There are multiple methods to measure AVR. For this study, Stokes' method was used to measure AVR because it accounts for the three-dimensionality of vertebra. Stokes' method offers a measurement of the axial rotation of the vertebrae by measuring the projected distances of the center of both pedicles from the center of the vertebra, accounting for vertebra width and level.[136] Using MIAS, AVR was calculated for each patient.[136]

3.7.6 Coronal Balance

Coronal balance is the horizontal distance between a vertical line drawn from the center of C7 and a second vertical line drawn from the center of S1.[137]

3.8 Statistical Analysis

3.8.1 Sample Description

Descriptive statistics related to demographic data (related to age and treatment) are summarized in the results section. Descriptive analysis was conducted using Statistical Package for the Social Sciences (SPSS) version 24.0 (SPSS, Inc., Chicago, IL, USA). For continuous data, average and standard deviation are presented.

The following statistical analyses were used:

- The Intra-class Correlation Coefficient ($ICC_{3.1}$) was used to examine test-retest reliability. Reliability analysis used data collected at 1 and 2 weeks following the initial the clinic visit. Absolute reliability was reported using the standard error of measurement (SEM).[78] Minimum detectable change (MDC) was calculated using the following formula: $MDC95 = SEM \times 1.96 \times \sqrt{2}$, whereby 1.96 corresponds to the value from the z distribution for a 95% confidence level.[78]

- Pearson correlation coefficients were used to evaluate the correlation between questionnaire scores and each radiographic measurement. A one-tailed significance level was used for hypothesis testing given the assumption that larger radiographic measurements are related to poorer quality of life and more negative perception of self-image.[94,95]

- Ceiling and floor effects were quantified for each score by calculating the percentage of participants obtaining the best and worst possible scores, respectively.[132]

3.9 Results

For the one-hundred participants that were recruited for this study, all were female (n = 100), had a mean age of 13.9±1.8 years, and a mean curve angle of 29º ±14º. Sixty-three (n = 63) received conservative treatment whereby 38 had a brace and 25 received exercise treatment. The remaining 37 participants were under clinical observation. Twenty participants (n = 20) had a mean upper thoracic curve of 19º ±11º, 99 participants had a mean thoracic curve of 24±18º, and 98 participants had a mean lumbar curve of 18º±16º. Participants in the reliability study had lower curve angles where the mean was 25º±9º (**Table 3-1).** The mean QOL as indicated by the SRS-22r Total score was 4.2±0.5. Means and standard deviations of other domains and of the SAQ versions are available in **Table 3-2**.

3.9.1 Test-retest Reliability

The scores of the multiple domains of the SAQ had lower test-retest reliability ($ICC_{3,1}$= 0.72 to 0.94) compared to the SAQ v1.1 for the Total, appearance, and expectations scores presented as $ICC_{3,1}$ (95%CI) of 0.92 (0.87; 0.96), 0.94 (0.89; 0.97), and 0.86 (0.75; 0.92), respectively. The Total score of the SRS-22r demonstrated high test-retest reliability, presented as $ICC_{3.1}$ (95%CI) = 0.94 (0.90 - 0.97) (**Table 3-3).**

3.9.2 Convergent Validity for SAQ Versions Against the SRS-22r Questionnaire

The SAQ domains scores had significant correlation with the SRS-22r Total score. The following is a breakdown of each SAQ domain score as they correlate with the SRS-22r Total score: General

(r = -0.59), Curve (r=-0.32), Prominence (r = -0.36), Trunk shift (r = -0.40), Waist (r = -0.40), Shoulders (r = -0.35), Kyphosis (r = -0.35), and Chest (r = -0.52) **(Table 3-4).** A significant correlation was also demonstrated between these SAQ domains and the SRS-22r domains (Total and Self-image) where r ranged between -0.21 and -0.70. The highest correlation was found between the SAQ general domain (r = -0.70) and SRS-22r self-image domain.

In contrast, the SAQ v1.1 Total and domains scores demonstrated a significant correlation with the SRS-22r Total score (r = -0.44 to -0.50). However, significant but lower correlation was found between the SAQ v1.1 Appearance and Expectation scores and the SRS-22r self-image score (r = -0.37 and -0.46). Additionally, a correlation was found between these SAQ v1.1 domains or total score and all SRS-22r domains (r= -0.23 to -0.50). (**Table 3-4**)

3.9.3 Convergent Validity for SAQ Versions Against the Radiographic Measurements

The Total score of the SAQ v1.1 had a significant correlation with the maximum Cobb angle, thoracic rotation, and coronal balance (r = 0.56, 0.40, and 0.29, respectively). Correlation of the SAQ v1.1 domain scores and radiograph measurements ranged from r = 0.12 to 0.63. Correlation of the SAQ domain scores ranged from 0.32 to 0.59 with the Cobb angle, and from 0.26 to 0.43 with thoracic rotation. All SAQ domains except General and Shoulders scores correlated with Coronal balance (|r|=0.25 to 0.35) A lower and non-significant correlation was observed between all domains of both SAQ versions and lumbar rotation except for the SAQ curve domain which had a significant correlation measuring r = 0.37 (**Table 3-5**).

3.9.4 Ceiling and Floor Effects

Ceiling and floor effects were observed only for the following domains of the SAQ: Curve= 11% (ceiling effect), Kyphosis= 68% (ceiling effect), and Waist= 4% (floor effect). Ceiling effects for the SAQ v1.1 domains were as follows: Total= 3%, Appearance= 5%, and Expectations = 14% (**Table 3-6**). Only the Expectations domain of the SAQ v1.1 presented floor effects at 14%.

3.10 Discussion

Only three domains of the SAQ (General, Prominence, and Chest) met the test-retest reliability threshold for research and individual use (ICC ≥ 0.90). The remaining domains met the threshold for research use only (ICC ≥ 0.70). In contrast and as hypothesized, the SAQ v1.1 Total score and Appearance scores have an adequate test-retest reliability that meets the threshold for use in research (ICC≥ 0.70) and for use on individuals (ICC≥0.90) per COSMIN. It is worth noting that the Expectations domain only met the threshold for use in research. This lower reliability might be attributed to the lower number of items (n = 4) in the Expectations domains compared to the Appearance domain (n = 10).

Evidence of adequate convergent validity was observed for both questionnaires. However, when examining the correlation of the SAQ scores with the SRS-22r Total score and Self-image domain, we observed that not all of the domains in the SAQ met the minimum threshold for convergence (r ≥ 0.35). In contrast, all domains of the SAQ v1.1 met the convergence threshold as initially hypothesized, indicating convergence validity for the SAQ v1.1.

We also hypothesized that the larger the radiograph measurements, the more negative the perceived self-image. Convergence with Cobb angle, an indicator of the scoliosis severity, was not found with all of the domains in the SAQ. In contrast, all domains of the SAQ v1.1 demonstrated levels of convergence, as we originally hypothesized. The Cobb angle had the highest correlation with the Appearance domain ($r = 0.63$). Thoracic vertebral rotation demonstrated convergence with the Appearance domain and Total score of the SAQ v1.1 but did not show the hypothesized levels of convergence with the majority of the SAQ domains. Coronal balance demonstrated convergence with only the SAQ v1.1 Appearance domain which can be, also, justified by the items imbalance mentioned earlier.

No significant correlation was found between the lumbar rotation and any of the domains except the Curve domain of the SAQ. This may be attributed to the low number of participants with a lumbar curve. Alternatively, the soft tissue surrounding the lumbar spine may deform and mask the underlying spine deformity from scoliosis, thereby decreasing the effect of the lumbar curve on the perceived self-image. Furthermore, the SAQ v1.1 has a higher correlation coefficient with the Cobb angle (an objective outcome measure) than the SRS-22r Self-image domain and the SRS-22r Total score (a self-reported subjective outcome measure). This is consistent with our hypothesis and the findings of the SAQ v1.1 development study.[88]

These results provide support on the appropriateness of the SAQ v1.1 for clinical and research use for assessing patients with AIS. This finding is consistent with previous studies. Carreon et al.[88] found the test-retest reliability of the SAQ v1.1 Appearance, Expectations, and Total domains to be $ICC_{3,1}$= 0.81, 0.91, and 0.89 which is consistent with our findings. Additionally, they found a

significant correlation between the SAQ v1.1 Appearance, Expectations, and Total scores to the SRS-22r Self-image score (r = -0.39, -0.32, and -0.43, respectively), SRS-22r Total score (r = -0.32, -0.21, and -0.34, respectively), and the Cobb angle (r = 0.36, 0.15, 0.32, respectively). This supports convergent validity. Findings from another study[77] showed convergence between the SAQ v1.1 Appearance, Expectations, and Total scores with the SRS-22r Self-image score (r= -0.39, -0.32, and -0.43, respectively) and the SRS-22r Total score (r= -0.32, -0.21, and -0.34, respectively) in a mixed sample of adolescents and adults with idiopathic scoliosis.

The SAQ v1.1 appears to be better at detecting changes in patients with AIS compared to the SAQ due to its domains having less than a 15% ceiling and floor effect. A study by Schreiber et al. shows that the SAQ has a ceiling effect of >27%.[42] The SAQ v1.1 shows promising ability to detect changes, has acceptable levels of ceiling effect, and presents the smallest SEM and MDC compared to the SAQ. Although the Expectations domain of the SAQ v1.1 has acceptable ceiling effect, it was the highest among other scores. This, however, may be attributed to the lower number of items in this domain compared to the Appearance domain. To reduce the ceiling effect, COSMIN suggests the need to add more items to the SAQ v1.1 expectations domain.[132]

3.10.1 Strengths and Limitations

Although this study is the first to examine and compare both the convergence validity and reliability of the SAQ and SAQ v1.1, it has some limitations. Namely, the recruited sample included adolescents with scoliosis who came from a single, tertiary hospital which may potentially limit the generalizability of our results. This could have reduced the variability of our

sample and, consequently, resulted in underestimation of reliability and overestimation of convergent validity for both questionnaires. Furthermore, the generalizability of our results to other age groups (such as adults and children with scoliosis), curve severity (i.e., >25°), and male adolescents with scoliosis may be limited given that our sample included only adolescent females with mild curve severity (i.e. 25 – 40°).

3.10.2 Future Directions

Future studies are recommended to evaluate the reliability, validity (e.g., construct validity and cross-cultural validity), and the responsiveness (i.e. the ability of the questionnaires to detect change)[132] of the SAQ v1.1 in a more broadly representative sample. These studies may use advanced statistical methods (e.g., the generalizability theory for reliability and multivariate analysis for validity) to fully understand the measurement properties of the SAQ v1.1 while accounting for possible sources of score variations.[78]

3.10.3 Conclusion

In conclusion, we support the use of the SAQ v1.1 over the SAQ in research and clinical practice for evaluating patients with AIS as the SAQ v1.1 has more adequate measurement properties.

Table 3-1: Description of the Participants

	Validity sample			Reliability sample		
	n	Range	Mean (SD)	n	Range	Mean (SD)
Age (years)	100	10 - 18	13.9 (1.8)	42	10 - 17	13.5 (1.8)
Maximum Cobb angle (°)	99*	10 - 74	29 (14)	41*	10 - 51	25 (9)
Maximum Thoracic rotation (°)	95	0.7 – 23.3	6.9 (4.3)	39	0.7 – 13.1	5.8 (3.4)
Maximum Lumbar rotation (°)	82	1.0 – 20.0	7.6 (4.0)	35	1.3 – 16.2	7.1 (3.2)
Coronal balance (mm)	99	0 – 22.1	7.2 (5.4)	41	0.2 – 22.0	7.2 (5.5)

n: Number of participants; **SD:** Standard deviation; **°:** Degree; *One image not available

Table 3-2: Means and Standard Deviation for All the Questionnaires at All Time Points

Questionnaire	Good QOL	Domains	Mean (SD)		
			Baseline (n= 100)	One week (n= 42)	Two weeks (n= 42)
SAQ	Low	General /5	7.4 (3.1)	6.6 (3.2)	6.3 (3.0)
		Curve /5	2.2 (0.7)	1.9 (0.5)	1.8 (0.4)
		Prominence /5	3.3 (1.2)	3.2 (1.3)	3.1 (1.2)
		Trunk shift /5	3.4 (1.2)	3.3 (1.2)	3.1 (1.0)
		Waist /5	6.6 (3.7)	5.8 (3.2)	5.6 (3.3)
		Shoulders /5	4.5 (2.1)	4.3 (2.0)	4.3 (2.1)
		Kyphosis /5	1.4 (0.7)	1.5 (0.7)	1.4 (0.6)
		Chest /5	3.7 (2.5)	3.6 (2.3)	3.6 (2.5)
SAQ v1.1	Low	Appearance /50	17.6 (5.3)	17.2 (5.1)	16.2 (4.3)
		Expectations /20	10.7 (5.4)	9.4 (4.8)	9.3 (5.3)
		Total /70	27.2 (9.2)	25 (8.4)	25.5 (8.6)
SRS-22r	High	Function /5	4.5 (0.5)	4.6 (0.4)	4.6 (0.4)
		Pain /5	4.2 (0.8)	4.3 (0.7)	4.3 (0.7)
		Self-image /5	4.1 (0.7)	4.0 (0.7)	4.1 (0.8)
		Mental health /5	3.9 (0.7)	4.0 (0.7)	4.1 (0.6)
		Satisfaction /5	3.9 (1.0)	4.2 (1.0)	4.2 (0.9)
		Total /5	4.1 (0.5)	4.2 (0.5)	4.3 (0.5)

n: Number of participants**, SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ:** Spinal Appearance Questionnaire**, SD:** Standard deviation, **QOL:** Quality of life.

Table 3-3: Test-retest Reliability of the Questionnaire Scores

Questionnaire	Good QOL	Domains	Test-retest reliability one vs. two weeks			
			N	ICC $_{3,1}$ (95% CI)	SEM (% of score)	MDC95 (% of score)
SAQ	Low	General /5	42	0.91 (0.84; 0.95)	0.96 (19.2)	2.66 (53.2)
		Curve /5	42	0.79 (0.64; 0.88)	0.22 (4.4)	0.61 (12.2)
		Prominence /5	42	0.90 (0.83; 0.95	0.35 (7)	0.96 (19.2)
		Trunk shift /5	42	0.89 (0.80; 0.94)	0.33 (6.6)	0.92 (18.4)
		Waist /5	42	0.72 (0.53; 0.84)	1.73 (34.6)	4.79 (95.8)
		Shoulders /5	42	0.85 (0.73; 0.91)	0.83 (16.6)	2.29 (45.8)
		Kyphosis /5	42	0.82 (0.69; 0.90)	0.28 (5.6)	0.77 (15.4)
		Chest /5	41	0.94 (0.89; 0.97)	0.58 (11.6)	1.61 (32.2)
SAQ v1.1	Low	Appearance /50	42	0.94 (0.89; 0.97)	1.06 (2.1)	2.95 (5.9)
		Expectations /20	42	0.86 (0.75; 0.92)	1.90 (9.5)	5.27 (26.4)
		Total /70	42	0.92 (0.87; 0.96)	2.3 (3.29)	6.32 (9.0)
SRS-22r	High	Function /5	42	0.84 (0.73; 0.91)	0.16 (3.2)	0.45 (9)
		Pain /5	42	0.96 (0.92; 0.98)	0.15 (3)	0.43 (8.6)
		Self-image /5	42	0.91 (0.83; 0.95)	0.23 (4.6)	0.63 (12.6)
		Mental health /5	42	0.91 (0.84; 0.95)	0.19 (3.8)	0.53 (10.6)
		Satisfaction /5	42	0.88 (0.79; 0.93)	0.33 (3.6)	0.91 (18.2)
		Total /5	42	0.94 (0.90; 0.97)	0.12 (2.4)	0.33 (6.6)

N: Number of participants**, SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ:** Spinal Appearance Questionnaire**, ICC:** Intra-class Correlation Coefficient**, SEM:** Standard Error of Measurement**, MDC95:** Minimal Detectable Change estimated using 95% Confidence Interval**, CI:** Confidence Interval**, QOL:** Quality of life

Table 3-4: Correlation Estimates for Convergent Validity between the Two SAQ Versions and the SRS-22r Scores

Questionnaire	Domain		SRS-22r					
			Function	Pain	Self-Image	Mental Health	Satisfaction	Total
SAQ	General	r	-.40**	-.32**	-.70**	-.43**	-.33**	-.59**
		n	100	100	100	100	99	100
	Curve	r	-.28**	-.24*	-.26*	-.19**	-.24*	-.32**
		n	100	100	100	100	99	100
	Prominence	r	-.30**	-.39**	-.26**	-.13**	-.30**	-.36**
		n	100	100	100	100	99	100
	Trunk shift	r	-.33**	-.34**	-.34**	-.24*	-.28**	-.40**
		n	100	100	100	100	99	100
	Waist	r	-.36**	-.39**	-.43**	-.24*	-.28**	-.40**
		n	100	100	100	100	99	100
	Shoulders	r	-.31**	-.30**	-.35**	-.13**	-.21*	-.35**
		n	100	100	100	100	99	100
	Kyphosis	r	-.37**	-.32**	-.21**	-.13**	-.29**	-.35**
		n	99	99	99	99	98	99
	Chest	r	-.44**	-.35**	-.51**	-.34**	-.30**	-.52**
		n	99	99	99	99	98	99
SAQ v1.1	Appearance	r	-.42**	-.41**	-.37**	-.24*	-.37**	-.48**
		n	100	100	100	100	99	100
	Expectations	r	-.34**	-.32**	-.46**	-.23*	-.31**	-.44**
		n	100	100	100	100	99	100
	Total	r	-.42**	-.40**	-.46**	-.26**	-.38**	-.50**
		n	100	100	100	100	99	100

n: Number of participants 100 unless specified, **r:** Pearson correlation coefficient, **SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ:** Spinal Appearance Questionnaire**.**

Table 3-5: Correlation Between SAQ Domain Scores from Each Version and Radiographic Measurements

Questionnaire	Domain		Maximum Cobb angle	Maximum Thoracic Rotation	Maximum Lumbar Rotation	Coronal balance
SAQ	General	r	.44**	.33**	.16	.20
		n	99	95	82	99
	Curve	r	.56**	.30**	-.37**	-.29**
		n	99	95	82	99
	Prominence	r	.51**	.34**	.10	.33**
		n	99	95	82	99
	Trunk shift	r	.59**	.43**	.16	.35**
		n	99	95	82	99
	Waist	r	.38**	.26*	.14	.25*
		n	99	95	82	99
	Shoulders	r	.40**	.30**	.05	0.08
		n	99	95	82	99
	Kyphosis	r	.47**	.31**	-.03	.31**
		n	98	94	81	98
	Chest	r	.32**	.40**	.04	.26*
		n	98	94	81	98
SAQ v1.1	Appearance	r	.63**	.46**	.19	.35**
		n	99	95	82	100
	Expectations	r	.38**	.25*	.12	.17
		n	99	95	82	99
	Total	r	.56**	.40**	.17	.29**
		n	99	95	82	99

n: Number of participants, **r:** Pearson correlation coefficient, **SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ:** Spinal Appearance Questionnaire**. QOL:** Quality of life

** Correlation is significant at the 0.01 level (2-tailed).

* Correlation is significant at the 0.05 level (2-tailed).

Table 3-6: Ceiling and Floor Effects for Each Questionnaire Score

Questionnaires	Good QOL	Domains		Ceiling (%)	Floor (%)
SAQ	Low	General	/5	0	0
		Curve	/5	11	0
		Prominence	/5	0	0
		Trunk shift	/5	0	0
		Waist	/5	0	4
		Shoulders	/5	0	0
		Kyphosis	/5	68	0
		Chest	/5	0	8
SAQ v1.1	Low	Appearance	/50	5	0
		Expectations	/20	14	10
		Total	/70	3	0

SAQ: Spinal Appearance Questionnaire**, QOL:** Quality of life

4 CHAPTER 4 – TEST-RETEST RELIABILITY AND CONVERGENT VALIDITY OF THE ISYQOL, TAASQ, AND BIDQ-S QUESTIONNAIRES IN ADOLESCENTS WITH IDIOPATHIC SCOLIOSIS

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Abstract

Introduction: Adequate measurement properties are a prerequisite for studies evaluating the effects of scoliosis treatments. New quality of life questionnaires including the Italian Spine Youth Quality Of life (ISYQOL), Truncal Anterior Asymmetry Scoliosis Questionnaire (TAASQ), and Body Image Disturbance Questionnaire-Scoliosis version (BIDQ-S) have recently been developed to address limitations of existing tools, but measurement properties for these new questionnaires should be compared.

Objective: This study aimed to determine the test-retest reliability and the convergent validity of the ISYQOL, TAASQ, and BIDQ-S questionnaires against radiographic measurements and established quality of life questionnaires such as the Scoliosis Research Society – 22 refined (SRS-22r) and Spinal Appearance Questionnaire version 1.1 (SAQ v1.1) in adolescents with idiopathic scoliosis.

Study Design: A metrological study was conducted using a cross-sectional design for validity, and a single-group, two repeated-measures design was used to test for reliability.

Sample: One-hundred females (n=100) with AIS with an age range of 10-18 years and who received non-operative treatment, were recruited from a Canadian pediatric scoliosis clinic.

Outcome Measures: Two established questionnaires (SRS-22r) and the (SAQ v1.1) and three new questionnaires (the ISYQOL, TAASQ, and BIDQ-S) were collected along with radiographic

measurements (maximum Cobb angle, coronal balance, and thoracic and lumbar vertebral rotations).

Methods: Questionnaires were administered electronically with a one-week interval between.

Results: Participants included 100 females with a mean age of 13.9±1.8 years and a curve angle of 28.8º ±13.9º. Test-retest reliability estimate ($ICC_{3,1}$) for new questionnaires ranged from 0.77 to 0.95. Convergence was demonstrated between the ISYQOL and SRS-22r (r = 0.67) and SAQ v1.1 (r = -0.53) Total scores. Correlations between the TAASQ domains for Breast, Appearance, and Clothing scores and the SAQ v1.1 Total score, Appearance domain, and Expectations domain were between r = -0.44 and -0.69. The domains of the TAASQ correlated with the SRS-22r Self-image and Total scores (r = 0.42 to 0.70). The convergence of the BIDQ-S was supported by a high correlation to the SRS-22r Total score (r = -0.65), SAQ v1.1 Total score (r = 0.62), SRS-22r Self-image domain (r = -0.55), and the SAQ v1.1 Appearance domain (r=0.60). All questionnaires presented ceiling or floor effects of ≥15% except for the ISYQOL. The ISYQOL score had a significant correlation only with the maximum Cobb angle (r = -.33). A significant correlation was found between the main domains of the TAASQ (Breast, Appearance, and Clothing) with the maximum Cobb angle, the thoracic vertebral rotation, and coronal balance (r= -0.20 to -0.47) but the correlation between the Clothing domain and coronal balance was not significant (r = -.13). The BIDQ-S showed a significant correlation with the maximum Cobb angle, the thoracic vertebral rotation, and coronal balance (r= 0.35, 0.33, and 0.23, respectively).

Conclusion: All new questionnaires demonstrated convergent validity and adequate reliability for research use in adolescent with scoliosis. Further, four domains of the TAASQ (Appearance, Clothing, Clothing General, and Clothing Specific) can be used in both research and clinical care in adolescents with idiopathic scoliosis.

4.1 Introduction

Adolescent idiopathic scoliosis (AIS) is the most common form of scoliosis for those whose age range from 10 to 18.[138] The prevalence of AIS is between 2-3% in this age group.[20] AIS corresponds to nearly 80% of all spine deformity cases for adolescents and is defined as "a structural, lateral, rotated curvature of the spine that arises in otherwise healthy children at or around puberty".[15, 21] Despite decades of research, the etiology of AIS remains unknown.

With AIS, the risk of scoliosis-related health problems developing into adulthood increases, resulting in decreased quality of life, cosmetic deformity, pain, and progressive functional limitations, especially if the curve magnitude is 30° or more at skeletal maturity.[26] If the curve is over 50°, it is almost certain that the curve will progress into adulthood, leading to deterioration in health-related quality of life. [26]

Health-related quality of life is a measure on the effects of a disease or a treatment on the physical, psychological, and social domains of functioning and well-being for a patient.[55] However, current outcome measures for *health-related quality of life* for patients with AIS show limitations in their measurements properties. The Scoliosis Research Society (SRS-22) questionnaire was not developed with input from patients and was initially created to measure QOL in candidates for surgery (i.e. patients with severe scoliosis). [132,138,139] In a study of 173 adolescent females with AIS treated conservatively, nine items of the SRS-22 had major ceiling effects (≥50%), (high proportion of participants with the best possible score), and 11 had moderate ceiling effects (≥20%).[139] Therefore, the SRS-22r may not accurately assess patients with milder forms of

scoliosis. Additionally, the SRS-22 was not developed using modern scaling techniques such as Rasch analysis.[132]

The Spinal Appearance Questionnaire (SAQ) assesses the perception patients have regarding their own appearance, as viewed from the back. This is not only impractical, but this view may also be of lesser importance to the patient compared to their appearance as viewed from the front.[85] Furthermore, the SAQ was also not developed with Rasch scaling, which ensures that the scores for the items are equally distributed.[79] This ensures that a measure has the attributes of being a continuous measurement, theoretically providing more power in analyses and the ability to detect change.[132] Lastly, the SAQ was not developed with patient input, which may therefore affect content validity.[85] Recently, Carreon et al.[88] developed a revised version of the SAQ named the Spinal Appearance Questionnaire version 1.1 (SAQ v1.1). Using factor analysis, they revised the SAQ into 14 items loaded onto two factors. The sum of the first 10 items make up the Appearance domain, and the sum of the last 4 items make up the Expectations domain. In contrast to the original version of the SAQ, the SAQ v1.1 has a total score that may facilitate a comparison between patients' perceived appearance in studies using different tools.

New questionnaires have been developed to address limitations of the SRS-22 and the SAQ. The Italian Spine Youth Quality of Life (ISYQOL) questionnaire was developed using Rasch analysis, and items on the ISYQOL were developed with input from patients, families, and clinicians to accurately assess changes related to treatment, ranging from conservative to surgical, for patients with varying levels of disease. [81] The input from patients and families is important to ensure that the items of the questionnaire are comprehensive and related to actual patient concerns. The

ISYQOL was translated to English from Italian using consensus provided by four team members. This translated ISYQOL was then reviewed by three Italian collaborators for compatibility with the original version.[83] The Italian collaborators found the English version to adequately represent the Italian version. The internal consistency of the ISYQOL has been tested in adolescents with idiopathic scoliosis and meets recommended internal consistency standards (α= 0.79 – 0.84).[83]

The Body Image Disturbance Questionnaire-Scoliosis version (BIDQ-S) is a short, 7-question survey that includes questions related to anterior appearance.[89] The Truncal Anterior Asymmetry Scoliosis Questionnaire (TAASQ) is an instrument that was recently developed to evaluate patient-endorsed concerns related to their anterior trunk appearance and the effects of these concerns on psychological perceptions and behavior.[140] In adolescents with idiopathic scoliosis, the BIDQ-S showed internal consistency (Cronbach alpha =0.82).[91]

Given the problems with existing measurements such as the SRS-22r and the SAQ v1.1, there is a need to assess the measurement properties of these new outcome measures. Such analysis may help determine whether these new measures adequately resolve the limitations presented in existing measurements used for patients with AIS. Additionally, metrological evidence may aid in choosing the best measures for assessing efficacy of AIS operative and non-operative interventions in both clinical practice and research.

4.2 Objective

This study aims to determine the test-retest reliability, convergent validity, ceiling and floor effects of the ISYQOL, TAASQ, and BIDQ-S questionnaires when compared against radiographic

measurements (maximum Cobb angle, coronal balance, and thoracic and lumbar vertebral rotations) and established quality of life questionnaires (SRS-22r and SAQ v1.1) in patients with AIS.

4.3 Study Design

A metrological study was conducted using a cross-sectional design for validity and ceiling and floor effects. Within a single-group, measures were repeated twice one week apart to assess test-retest reliability.

4.4 Hypothesis

We hypothesized that:

1. The test-retest reliability of the new questionnaires will be adequate and meet the minimum acceptable standards suggested by COSMIN for measurement for use in groups of patients (ICC ≥ 0.70) and for use in individual patients (ICC ≥ 0.90).[132]

2. The ISYQOL scores indicating better QOL will have correlation coefficients ≥ 0.35 with better QOL scores on the SRS-22r, better-perceived appearance on the SAQ v1.1, and smaller radiographic measurements.

3. The scores for the TAASQ and BIDQ-S questionnaires, both indicating better self-image, will have correlation coefficients ≥ 0.35 with a better-perceived appearance on the SAQ v1.1, SRS-22r self-image domain, and smaller radiographic measurements.

4. All questionnaires will be free from ceiling and floor effects or have less than 15% ceiling or floor effect.[132]

These hypotheses were based on the assumption that larger radiographic measurements has been related to poorer quality of life and more negative perception of self-image.[94,95] We expected stronger correlations among the tools or domains most focused on self-image/perceived appearance (such as the SAQ, SAQ v1.1 Appearance domain, BIDQ-S, TAASQ, and SRS-22r Self-image domain) and among the tools measuring overall QOL (such as the SRS-22r and ISYQOL).

4.5 Sample

Participants were recruited from the Stollery Children's Hospital while attending routine scoliosis clinic visits. Informed consent was obtained from eligible participants and their legal guardians. This study, including its consent documentation, was reviewed and approved by the University of Alberta Health Research Ethics Board (Pro00073569). A total of one-hundred participants (n=100) were recruited. *Inclusion criteria* for this study included: having an active diagnosis of idiopathic scoliosis, be of 10-18 years of age, have curve severity over 10°, and being fluent in English. *Exclusion criteria* included: having a history of spine surgery, having a history or active diagnosis of other diseases that might affect quality of life and/or torso appearance, or having history of trauma in the torso or lower extremities

4.6 Outcome Measures

4.6.1 Scoliosis Research Society-22 Refined Questionnaire (SRS-22r)

The Scoliosis Research Society-22 refined questionnaire (SRS-22r) is the most commonly used instrument for measuring HRQOL in patients with scoliosis.[75] It consists of 22 questions covering five domains: pain, self-image, function, mental health, and satisfaction with management. Scores range from 1 to 5 per question, and these scores are averaged to produce domain scores and a total score.[75] Higher scores on the SRS-22r indicate better quality of life. The SRS-22 demonstrated acceptable reliability (ICC = 0.85 to 0.96) and concurrent validity ($r > 0.70$) compared to the Short Form 36 (SF-36). However, 56.9% of respondents were given the highest score possible, showing that this assessment has potential ceiling effects and floor effects for patients with post-operative idiopathic scoliosis.[75] The SRS-22 has been refined to improve the internal consistency of its function domain, increasing to α=0.67 to 0.78 following changes.[134]

4.6.2 Spinal Appearance Questionnaire Version 1.1 (SAQ v1.1)

The Spinal Appearance Questionnaire version 1.1 (SAQ v1.1) is a modified version of the SAQ. It consists of 14 items. The first ten items are related to the 'Appearance' domain and the last four items are related to the 'Expectations' domain. The ten items related to the appearance domain are as follows: body curve, rib prominence, flank prominence, head-chest-hips, position of head over hips, shoulder level, shoulder blade rotation, shoulder angle, head position, and spine prominence. The four items related to the expectations domain are as follows: 'I want to be more even'; 'I want to have more even shoulders'; 'I want to have more even hips', and 'I want to have a more even waist'. For the 'Appearance' domain, response options are provided as drawings ranging from

mild (score of 1) to severe (score of 5). The 'Expectations' domain features response options ranging from 'not true' (score of 1) to 'very true' (score of 5). Answers are subsequently summed to provide a score ranging from 10 to 50 for the 'Appearance' domain and ranging from 4 to 20 for the 'Expectations' domain. The SAQ v1.1 total score is calculated by summing the total scores of both domains.[88] The best possible total score for the SAQ v1.1 is 14, related to better QOL and more positive perception of scoliosis related deformity, and the worst possible score is 70.[88] In a study conducted by Carreon and colleagues, 1,802 adolescents with AIS were given the SAQ v1.1 and their results suggested that the SAQ v1.1 had good reliability (Cronbach's $\alpha \geq 0.88$; test-retest correlation ≥ 0.81) and convergent validity as it relates to major curve magnitude ($r=0.32 – 0.36$).[88]

4.6.3 Italian Spine Youth Quality of Life Questionnaire (ISYQOL)

The ISYQOL is a new patient reported outcome measurement (PROM) for health-related quality of life in adolescents with idiopathic scoliosis.[12] The ISYQOL aims to accurately assess changes related to treatment, ranging from conservative to surgical, for patients with varying levels of disease.[12] Items of the ISYQOL were generated following a content analysis of the concerns expressed by patients, parents, and scoliosis specialists from an online forum.[12] The ISYQOL was developed using Rasch analysis, which ensures that the items are equally distributed and scores can serve as a continuous measurement.[12] Studies analyzing the measurement properties of the ISYQOL are promising. For example, Caronni and colleagues concluded that the ISYQOL performs better than the SRS22, having better validity and ability to detect the impact of disease severity on health-related quality of life.[23]

4.6.4 Truncal Anterior Asymmetry Questionnaire (TAASQ)

The TAASQ was recently developed to assess patient-perceived anterior truncal appearance.[140] This consists of 14 individual questions that fall into three main domains: Breast (questions 4 a, b, c; 12; 13; 14), Appearance (5; 6; 7; 10; 11), and Clothing (1; 2 a, b, c, d, e; 3; 8 a, b, c; 9). The Clothing domains can be further classified into two Clothing subdomains: Clothing-General (Q1, 3, 8a, 9) and Clothing-Specific (Q2, 8b, 8c, 8d). Furthermore, the Breast domain can be categorized into three subdomains: Breast location (Q4c, 13), Breast Shape (Q4b, 12), and Breast size (Q4a, 14). The TAASQ uses a combination of Likert scales and free text responses, making up 22 scored items. Each multiple-choice question features answer options ranging from 1 to 5, where 5 corresponds to the least amount of concern or asymmetry and 1 corresponds to the most concern. The average score is then calculated for each domain and sub-domain. Higher scores on the TAASQ indicate better quality of life.[140] Based on a review on current literature, we found that no studies have investigated the measurement properties of the TAASQ.

4.6.5 Body Image Disturbance Questionnaire - Scoliosis version (BIDQ-S)

The Body Image Disturbance Questionnaire - Scoliosis version (BIDQ-S) was derived from the generic BIDQ specifically for use by patients with scoliosis.[141] The BIDQ-S evaluates perceptions of body image disturbance and is used to identify patients who are experiencing distress and impairment related to their appearance concerns.[141] The BIDQ-S consists of 7 questions. Each answer, ranging from a response of 1 to 5, is scored. The total score is obtained by calculating the mean score of the seven items. Lower scores on the BIDQ-S indicate better quality of life. The

BIDQ-S demonstrates good internal consistency in patients with AIS (Cronbach alpha = 0.82) and convergent validity to the SRS-22 Total score (r = -0.50 to -0.72). [141]

4.6.6 Cobb Angle

The Cobb angle is a widely used measurement to evaluate scoliotic curve severity and risk for progression. It also helps direct brace and surgical treatment decisions in patients with scoliosis.[1] The Cobb angle is considered the most accurate and reliable method to quantify scoliosis severity, and a definitive diagnosis of scoliosis cannot be made without it. [138'15] To obtain the Cobb angle, a straight line was drawn along the upper endplate of the most tilted vertebra above the apex and along the lower endplate of the most tilted vertebra below the apex of each curve. The apex was identified by choosing the most translated vertebra in each of the spine curvatures. The angle between these two lines forms the Cobb angle.[24]

4.6.7 Axial Vertebral Rotation (AVR)

Axial vertebral rotation is the rotation of a vertebra around a vertical axis when projected onto the transverse image plane.[135] There are multiple methods to measure AVR. Stokes' method was used to measure AVR because it accounts for the three-dimensionality of vertebra. Stokes' method offers a measurement of the axial rotation of the vertebrae by measuring the projected distances of the center of both pedicles from the center of the vertebra, accounting for vertebra width and level.[136] Using custom software in Medical Image Analysis Software (MIAS) (version 9.6.7.0) Professional Edition, AVR for thoracic and lumbar curves was calculated for each patient.[136]

4.6.8 Coronal Balance

Coronal balance is the horizontal distance between a vertical line drawn from the center of C7 and a second vertical line drawn from the center of S1.[137]

4.7 Methods

Questionnaires were programmed in the Research Electronic Data Capture (REDCap) software and administered as an online survey (version 7.6.9). Using the automatic invitation feature in REDCap, participants completed questionnaires online at the clinic visit when the study was first introduced and one week and two weeks after this visit. Participants required roughly 15 minutes to complete all five questionnaires, which were presented in the following order: ISYQOL, SRS-22r, SAQ v1.1, BIDQ-S, and TAASQ. To minimize missing data, an automatic prompt appeared if any questions were skipped during response submission.

At the start of the study, standing posterior-anterior radiographs of the full spine were obtained using the EOS system. The Cobb angle, axial vertebral rotation, and coronal balance were subsequently measured by one rater (MA) using custom MIAS software (version 9.6.7.0). Prior to data collection, MA, a physical therapist, was trained to collect this data in a uniform manner by observing one demonstration, discussing ten practice cases, and conducting these measurements twice ($ICC_{3,1}$= 0.99, 95% CI .94 to 1).

Radiographic files were uploaded to MIAS after being converted from Digital Imaging and Communications in Medicine (DICOM) to Joint Photographic Experts Group (JPEG) image

format using ImageJ software (version 1.50i) (Rasband, W.S., ImageJ, U. S. National Institutes of Health, Bethesda, Maryland, USA, https://imagej.nih.gov/ij/, 1997-2018).

MIAS was also used to measure the coronal balance and AVR using the Stokes' method. The evaluator was blinded to the identity of the participants and the questionnaire scores during image measurement.

4.8 Statistical Analysis

4.8.1 Sample Description

Descriptive statistics related to demographic data (related to age and treatment) is summarized in the results section. Descriptive analysis was conducted using Statistical Package for the Social Sciences (SPSS) version 24.0 (SPSS, Inc., Chicago, IL, USA). Box plots for each variable were analyzed to understand score distribution. For continuous data, average and standard deviation were presented.

The following statistical analyses were performed:

- The Intra-class Correlation Coefficient ($ICC_{3.1}$) was used to examine test-retest reliability. Reliability analysis used data collected at 1 and 2 weeks following the initial the clinic visit. Absolute reliability was reported using the standard error of measurement (SEM).[78] Minimum detectable change (MDC) was calculated using the following formula: $MDC95 = SEM \times 1.96 \times \sqrt{2}$, whereby 1.96 corresponds to the value from the z distribution for a 95% confidence level.[78]

- Pearson correlation coefficients were used to evaluate the correlation between questionnaire scores and each radiographic measurement. A one-tailed significance level was used for hypothesis testing given the assumption that larger radiographic measurements are related to poorer quality of life and more negative perception of self-image.[94,95]

- Ceiling and floor effects were quantified for each score by calculating the percentage of participants obtaining the best and worst possible scores, respectively.[132]

Results related to the satisfaction domain of the SRS-22r were not analyzed because patients did not have a long history of care (many were being evaluated for their first visit)

4.9 Results

One-hundred participants were recruited for this study. All were female (n = 100), had a mean age of 13.9±1.8 years, and a mean curve angle of 29^{o} ±14^{o}. Sixty-three (n = 63) received conservative treatment whereby 38 had a brace and 25 received exercise treatment. The mean QOL as scored on the SRS-22r and SAQ v1.1was 4.13±0.53 and 27.17±9.15, respectively (**Table 4-1**). The characteristics of the 42 participants in the reliability study are reported in **Table 4-1.**

4.9.1 Test-retest Reliability

The Total score of the SAQ v1.1 and the SRS-22r demonstrated the highest test-retest reliability among all five questionnaires ($ICC_{3.1}$ [95%CI] = 0.92 [0.87 – 0.96] and 0.94 [0.90 - 0.97], respectively). The ISYQOL, BIDQ-S, and all of the TAASQ domain scores had test-retest $ICC_{3.1}$

of 0.77 or higher. The scores of the TAASQ Appearance and Clothing domains and all Clothing subdomains had ICCs exceeding 0.9. The percentage of the score scales corresponding to the SEM and MDC ranged between 8.6% and 26.4%. BIDQ-S had the lowest SEM and MDC among the new questionnaires (3.2% and 9%, respectively) whereas the highest SEM and MDC were observed for the TAASQ Breast size domain (6.2% and 17.4%, respectively) (**Table 4-3).**

4.9.2 Convergent Validity Estimates Between New and Established Questionnaires

The ISYQOL score had a significant correlation with the SRS-22r Total, function, pain, self-image, and mental health scores (r= 0.67, 0.49, 0.53, 0.53 and 0.48, respectively); it also had correlation with the SAQ v1.1 Total score (r=-0.53), Appearance (r=-0.46), and the Expectations domains (r=-0.50). (**Table 4-4**).

Scores of the main TAASQ domains (Breast, Appearance, and Clothing) correlated significantly with the SRS-22r Total score (r = 0.47, 0.65 and 0.60, respectively) and with the SRS-22r self-image domain (r = 0.42, 0.63 and 0.70, respectively). Scores of the main TAASQ domains (Breast, Appearance, and Clothing) significantly correlated with the SAQ v1.1 Total score (r = -0.53, -0.69 and -0.52, respectively). The correlation between the TAASQ Appearance domain and the SAQ v1.1 appearance domain was r = -0.63. All TAASQ subdomains correlated significantly with the SRS-22r Total score (r = 0.32 to 0.57) and SAQ v1.1 Total score (r= -0.36 to -0.55) (**Table 4-4**).

The BIDQ-S correlated significantly with the SRS-22r Total and the self-image domain (r = -0.65 and -0.55, respectively)**.** Additionally, the BIDQ-S correlated significantly with the SAQ v1.1

Total score (r = 0.62), Appearance domain (r = 0.60), and Expectations domain (r = 0.52) (refer to **Table 4-4**).

4.9.3 Convergent Validity Between New Questionnaires Radiograph Measurements

With the radiographic measurements, the ISYQOL score had a significant correlation only with the maximum Cobb angle (r = -0.33). A significant correlation was found between the main domains of the TAASQ (Breast, Appearance, and Clothing) and the maximum Cobb angle, the thoracic vertebral rotation, and coronal balance (r= -0.24 to -0.47). A low and non-significant correlation was observed between the Clothing domain and coronal balance (r = -0.13) (**Table 4-5**). The BIDQ-S showed a significant correlation with the maximum Cobb angle, the thoracic vertebral rotation, and coronal balance (r= 0.35, 0.33, and 0.23, respectively). None of the questionnaires correlated with lumbar rotation in this sample (**Table 4-5**).

4.9.4 Ceiling and Floor Effects

Only 2% of the participants scored the highest possible ISYQOL score and no participants scored the lowest possible score. The main domains of the TAASQ presented Floor effects (Breast= 39%, Appearance= 8%, and Clothing= 15%). For the BIDQ-S, 1% of participants scored the lowest, and 21% scored at the highest possible score. In contrast, the percentage of ceiling effects in the affected domains of the SRS-22r was as follow: Function= 19% and Pain= 18%. Ceiling effects for the SAQ v1.1 domains were as follows: Total score= 3%, Appearance= 5%, and Expectations= 14%. The Expectations domain of the SAQ v1.1 demonstrated a floor effect of 10% (**Table 4-6**).

4.10 Discussion

The scores of all questionnaires (domains and subdomains) that have been examined in this study (SRS-22r, SAQv1.1, ISYQOL, TAASQ, and BIDQ-S) demonstrated adequate test-retest reliability for research use (ICC≥0.70). The SRS-22r (Pain domain score, Self-image, Mental health and Total score), SAQ v1.1 (Appearance domain and Total score), and the TAASQ (Clothing domain, and the Appearance domain) also show adequate reliability for individual use (ICC≥0.90).

Overall, the ISYQOL, TAASQ (except for the correlation between SAQ v1.1 Expectations and Breast size domain), and BIDQ-S demonstrated convergent validity by significantly correlating with the SAQ v1.1 (Table 4-4). Similarly, the ISYQOL, TAASQ (except the Breast size domain), and BIDQ-S demonstrated convergent validity by significantly correlating with the SRS-22r (Table 4-4). Finally, while the TAASQ (except the Breast size and Clothing specific domains) and BIDQS showed correlation with radiographic measurements, the ISYQOL did not (Table 4-5).

To our knowledge, no previous studies have examined the reliability and validity of the TAASQ in adolescents with idiopathic scoliosis. However, a previous study concluded that the ISYQOL has better known-groups validity and is more sensitive at detecting impact of disease severity on HRQOL in adolescents with scoliosis compared to the SRS-22.[142] Additionally, our lab found the translated ISYQOL to meet the recommended standards for internal consistency in adolescents with idiopathic scoliosis (α= 0.79 - 0.84).[83] Similarly, the BIDQ-S has previously demonstrated good internal consistency in adolescents with idiopathic scoliosis who required surgery (Cronbach

alpha = 0.82). Also, the BIDQ-S demonstrated convergent validity with the SRS-22 domains and Total score (r = -0.42 to -0.65).[141]

Notably, the Breast domain of the TAASQ did not meet the reliability threshold for individual use (ICC≥0.90). we hypothesised that questionnaires with items focused on less relevant problems for the younger adolescent age group (such as breast shape, size, and location) may be less reliable. Thoracic vertebral rotation demonstrated convergence only with Breast, Breast shape, Appearance, and Clothing domains, which is logical given that thoracic curves are the most common type of curves in adolescents with idiopathic scoliosis and are more likely to affect breast appearance than any other changes reflected in our radiographic measurements.[143] Finally, no significant correlation was found between and the ISYQOL and the radiographic measurements. This can be explained by the nature of the ISYQOL which is a questionnaire assessing QOL, rather than self-image perceptions. It also appears that the ISYQOL was designed to capture minor changes in quality of life to maximize its ability to detect changes. This was demonstrated by the lack of notable ceiling effects in the present study. Therefore, it may capture changes before the radiographic severity is sufficient to affect function and be visible externally.

Together with prior reliability evidence for the ISYQOL[83], this study supports its use for QOL research in AIS. Additionally, the ISYQOL appears to be better at detecting changes compared to the SRS-22r and the SAQ V1.1 as its is free from ceiling and floor effects. However, longitudinal responsiveness studies are needed before recommending wider implementation of the ISYQOL because questionnaires with relatively large SEM and MDC (ISYQOL= 4.9% and 13.6%, respectively) have a limited ability to identify small changes as real changes.

The new questionnaires measuring a similar perceived appearance construct (such as the BIDQ-S and TAASQ) demonstrated an acceptable level of measurement reliability and convergent validity. They captured anterior perceived appearance with more relevance to patients than the SAQ v1.1 showed adequate reliability and validity. However, the BIDQ-S, TAASQ, and the SAQ v1.1 still suffer from ceiling or floor effects. Regardless, the BIDQ-S is a shorter assessment than the SAQ v1.1 and the TAASQ, has less ceiling effect than both, and presented the smallest SEM and MDC compared to the SAQ v1.1 and the TAASQ.

We support the use of all new questionnaires in research and the use of the TAASQ Appearance and Clothing domains and subdomains for individuals. However, researchers and clinicians seeking to use these instruments should weigh the following considerations. First, they should consider the limitations of each questionnaire. For instance, we should be careful when interpreting the scores of the questionnaires with ceiling effects. If patient responses are clustered at the extremes of a questionnaire's scale (ceiling or floor effect), the questionnaire's ability to detect change will be affected because there is no more room for detecting further improvement or deterioration, respectively.

The second factor is the context in which the measurement will be used. For example, in situations where time is limited, a rater might choose the BIDQ-S over other questionnaires as it is the shortest to administer. Lastly, one must take into consideration the decisions that will be made based on the results. For example, a measurement that will be used to discriminate between patients to determine who will undergo major surgery and those who will not, should have higher convergent validity since more serious clinical consequences hinge on the measurement results.

4.10.1 Strengths and Limitations

To our knowledge, this is the first study to evaluate the measurement properties of the new questionnaires against established questionnaires and this array of radiograph measurements. However, the generalizability of our results might be limited due to recruiting from a single specialized clinic and the exclusion of surgical candidates. However, we decided to limit our sample to patients treated conservatively because this is an understudied population and due to concerns that existing tools may not perform well in this population. Additionally, many of the participants (58%) did not participate in the retest. This is possibly due to questionnaire fatigue resulting from completing five questionnaires twice within a one-week interval. If reliability was related to interest in repeated administration, our results may not generalize to the whole population. Still, comparisons among questions were based on the same sample so that relative performance could be interpreted.

4.10.2 Future Directions

Future studies are recommended to evaluate responsiveness and to assess the ability of these questionnaires to detect change. Furthermore, we used the classical test theory (CTT) in this study, and future studies may use the generalizability theory to identify the source of score variation and item response theory to examine scaling issues. Additionally, future studies could evaluate how these questionnaires perform in different languages as well as in different age groups such as adults with scoliosis and patients with other spinal deformities.

4.10.3 Conclusion

All new questionnaires demonstrated convergent validity and adequate reliability for research use in adolescent with scoliosis. However, four domains of the TAASQ (Appearance, Clothing, Clothing General, and Clothing Specific) can be used in both research and clinical care in adolescents with idiopathic scoliosis. However, to inform selection, an outcome measure administrator should consider measurement limitations, the measurement context, and the decisions that will be made based on its result.

Table 4-1: Description of the Participants

	Validity sample			Reliability sample		
	n	Range	Mean (SD)	n	Range	Mean (SD)
Age (years)	100	10 -18	13.9 (1.8)	42	10 -17	13.5 (1.8)
Maximum Cobb angle (°)	99	10- 74.4	28.8 (13.9)	41	10 - 51	24.7 (9.3)
Maximum Thoracic rotation (°)	95	0.7 – 23.3	6.9 (4.3)	39	0.7 – 13.1	5.8 (3.4)
Maximum Lumbar rotation (°)	82	1.0 - 20	7.6 (4)	35	1.3 – 16.2	7.13 (3.2)
Coronal balance (mm)	99	0 – 22.1	7.2 (5.4)	41	0.2 – 22	7.2 (5.5)

n: Number of participants

SD: Standard deviation

°: Degree

Table 4-2: Means and Standard Deviation for All the Questionnaires at All Time Points

Questionnaire	Good QOL	Domains		Mean (SD)		
				Baseline (n= 100)	One week (n= 42)	Two weeks (n= 42)
SRS-22/5	High	Function	/5	0.52 (4.46)	4.57 (0.41)	4.60 (0.40)
		Pain	/5	4.18 (0.82)	4.31 (0.74)	4.32 (0.74)
		Self-image	/5	4.06 (0.66)	4.08 (0.73)	4.13 (0.77)
		Mental health	/5	3.90 (0.74)	4.02 (0.70)	4.05 (0.64)
		Satisfaction	/5	3.85 (0.96)	4.15 (0.95)	4.17 (0.93)
		Total	/5	4.13 (0.53)	4.24 (0.50)	4.26 (0.50)
SAQ v1.1	Low	Appearance	/50	17.57 (5.33)	17.15 (5.13)	16.21 (4.25)
		Expectations	/20	10.65 (5.40)	9.39 (4.80)	9.31 (5.26)
		Total	/70	27.17 (9.15)	25.4 (8.4)	25.52 (8.56)
ISYQOL	High	Spine health	/100	59.72 (12.38)	62.94 (14.14)	64.08 (14.84)
TAASQ	Low	Breast	/5*	4.60 (0.53)	4.59 (0.52)	4.62 (0.55)
		Breast location	/5*	4.77 (0.49)	4.76 (0.47)	4.73 (0.49)
		Breast shape	/5*	4.50 (0.64)	4.53 (0.57)	4.60 (0.58)
		Breast size	/5*	4.53 (0.72)	4.49 (0.70)	4.54 (0.74)
		Appearance	/5	4.10 (0.71)	4.21 (0.70)	4.21 (0.73)
		Clothing	/5	4.13 (0.70)	4.22 (0.75)	4.30 (0.75)
		Clothing general	/5	4.13 (0.73)	4.23 (0.80)	4.27 (0.87)
		Clothing specific	/5	4.21 (0.69)	4.30 (0.69)	4.40 (0.67)
BIDQ-S	Low	Total	/5	1.52 (0.63)	1.35 (0.47)	140 (0.48)

n: Number of participants**, ISYQOL:** Italian Spine Youth Quality of Life**, SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ v1.1:** Spinal Appearance Questionnaire version v1.1**TAASQ:** Truncal Anterior Asymmetry Scoliosis Questionnaire**, BIDQ-S:** Body Image Disturbance Questionnaire-Scoliosis version**, SD:** Standard deviation, **QOL:** Quality of life, **n*:** n=41.

Table 4-3: Test-retest Reliability of the Scores of the Questionnaires

	Good QOL	Domains		Test-retest reliability one vs. two weeks			
				N	ICC $_{3,1}$ (95 CI)	SEM (% of score)	MDC95 (% of score)
SRS-22	High	Function	/5	42	0.84 (0.73 - 0.91)	0.16 (3.2)	0.45 (9.0)
		Pain	/5	42	0.96 (0.92 - 0.98)	0.15 (3.0)	0.43 (8.6)
		Self-image	/5	42	0.91 (0.83 - 0.95)	0.23 (4.6)	0.63 (12.6)
		Mental health	/5	42	0.91 (0.84 - 0.95)	0.19 (3.8)	0.53 (10.6)
		Satisfaction	/5	42	0.88 (0.79 - 0.93)	0.33 (3.6)	0.91 (18.2)
		Total	/5	42	0.94 (0.90 - 0.97)	0.12 (2.4)	0.33 (6.6)
SAQ v1.1	Low	Appearance	/50	42	0.94 (0.89 - 0.97)	1.06 (2.1)	2.95 (5.9)
		Expectations	/20	42	0.86 (0.75 - 0.92)	1.90 (9.5)	5.27 (26.4)
		Total	/70	42	0.92 (0.87 – 0.96)	2.3 (3.3)	6.32 (9.0)
ISYQOL	High	Spine health	/100	42	0.89 (0.80 - 0.94)	4.9 (4.9)	13.60 (13.6)
TAASQ	Low	Breast	/5	41	0.84 (0.72 - 0.91)	0.22 (4.4)	0.61 (12.2)
		Breast location	/5	41	0.77 (0.60 - 0.87)	0.24 (4.8)	0.66 (13.2)
		Breast shape	/5	41	0.83 (0.70 - 0.90)	0.25 (5.0)	0.68 (13.6)
		Breast size	/5	41	0.82 (0.68 - 0.90)	0.31 (6.2)	0.87 (17.4)
		Appearance	/5	42	0.93 (0.87 - 0.96)	0.19 (3.8)	0.53 (10.6)
		Clothing	/5	42	0.95 (0.90 - 0.97)	0.17 (3.4)	0.46 (9.2)
		Clothing general	/5	42	0.91 (0.84 - 0.95)	0.24 (4.8)	0.67 (13.4)
		Clothing specific	/5	42	0.91 (0.85 - 0.95)	0.20 (4.0)	0.55 (11.0)
BIDQ-S	Low	Total	/5	42	0.89 (0.80 - 0.94)	0.16 (3.2)	0.45 (9.0)

n: Number of participants**, ISYQOL:** Italian Spine Youth Quality of Life**, SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ v1.1:** Spinal Appearance Questionnaire version v1.1**, TAASQ:** Truncal Anterior Asymmetry Scoliosis Questionnaire**, BIDQ-S:** Body Image Disturbance Questionnaire-Scoliosis version.**, ICC:** Intra-class Correlation Coefficient**, SEM:** Standard Error of Measurement**, MDC:** Minimal Detectable Change**, CI:** Confidence Interval**, QOL:** Quality of life

Table 4-4: Correlation Estimates for Convergent Validity Between the New and Established Questionnaires Scores

		SAQ v1.1			SRS-22r				
		Appearance	**Expectations**	**Total**	**Function**	**Pain**	**Self-Image**	**Mental Health**	**Total**
ISYQOL - Spinal Health	r	-.46**	-.50**	-.53**	.49**	.53**	.53**	.48**	.67**
	n	100	100	100	100	100	100	100	100
TAASQ – Breast	r	-.48**	-.47**	-.53**	.36**	.34**	.42**	.38**	.47**
	n	98	98	98	98	98	98	98	98
TAASQ - Breast location	r	-.39**	-.42**	-.45**	.41**	.36**	.44**	.41**	.50**
	n	98	98	98	98	198	98	98	98
TAASQ - Breast shape	r	-.52**	-.47**	-.55**	.36**	.29**	.38**	.30**	.42**
	n	98	98	98	98	98	98	98	98
TAASQ - Breast size	r	-.32**	-.33**	-.36**	0.19	.24*	.29**	.30**	.32**
	n	98	98	98	98	98	98	98	98
TAASQ - Appearance	r	-.63**	-.61**	-.69**	.44**	.45**	.63**	.50**	.65**
	n	100	100	100	100	100	100	100	100
TAASQ - Clothing	r	-.44**	-.50**	-.52**	.30**	.37**	.70**	.49**	.60**
	n	100	100	100	100	100	100	100	100
TAASQ-Clothing general	r	-.44**	-.47**	-.50**	.30**	.31**	.67**	.46**	.57**
	n	100	100	100	100	100	100	100	100
TAASQ -Clothing Specific	r	-.38**	-.47**	-.47**	.28**	.35**	.64**	.47**	.56**
	n	100	100	100	100	100	100	100	100
BIDQ-S Total	r	.60**	.52**	.62**	-.56**	-.46**	-.55**	-.43**	-.65**
	n	100	100	100	100	100	100	100	100

n: Number of participants, **r:** correlation coefficient, **ISYQOL:** Italian Spine Youth Quality of Life**, SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ v1.1:** Spinal Appearance Questionnaire version v1.1**, TAASQ:** Truncal Anterior Asymmetry Scoliosis Questionnaire**, BIDQ-S:** Body Image Disturbance Questionnaire-Scoliosis version**, SD:** Standard deviation, **QOL:** Quality of life.

Table 4-5: Correlation Between the Questionnaire Scores and the Radiograph Measurements.

	Good QOL	Maximum Cobb angle	Maximum Thoracic Rotation	Maximum Lumbar Rotation	Coronal balance
ISYQOL - Spinal health	High	-.33** (n=99)	-.26* (n=95)	-.05 (n=82)	-.08 (n=99)
TAASQ - Breast	Low	-.32** (n=97)	-.37** (n=94)	.09 (n=80)	-.36** (n=97)
TAASQ - Breast location		-.26** (n=97)	-.30** (n=94)	.12 (n=80)	-.34** (n=97)
TAASQ - Breast shape		-.39** (n=97)	-.45** (n=94)	.02 (n=80)	-.38** (n=97)
TAASQ - Breast size		-.16 (n=97)	-.20 (n=94)	.09 (n=80)	-.22* (n=97)
TAASQ - Appearance		-.47** (n=99)	-.44** (n=95)	-.17 (n=82)	-.24* (n=99)
TAASQ - Clothing		-.29** (n=99)	-.35** (n=95)	-.14 (n=82)	-.13 (n=99)
TAASQ - Clothing general		-.38** (n=99)	-.32** (n=95)	-.18 (n=82)	-.10 (n=99)
TAASQ - Clothing specific		-.20* (n=99)	-.31** (n=95)	-.11 (n=82)	-.12 (n=99)
BIDQ-S Total	Low	.35** (n=99)	.33** (n=95)	.20 (n=82)	.23* (n=99)

n: Number of participants**, ISYQOL:** Italian Spine Youth Quality of Life**, SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ v1.1:** Spinal Appearance Questionnaire version v1.1**, TAASQ:** Truncal Anterior Asymmetry Scoliosis Questionnaire**, and BIDQ-S:** Body Image Disturbance Questionnaire-Scoliosis version.

** Correlation is significant at the 0.01 level (2-tailed). * Correlation is significant at the 0.05 level (2-tailed). + Number of participants with lumbar curve ranged between 80 – 82.

Table 4-6: Ceiling and Floor Effects for the Questionnaire Scores

Questionnaires	Good QOL	Domains		Ceiling (%)	Floor (%)
SRS-22/5	High	Function	/5	19	0
		Pain	/5	18	0
		Self-image	/5	10	0
		Mental health	/5	8	0
		Satisfaction	/5	22	1
		Total	/5	0	0
SAQ v1.1/5	Low	Appearance	/50	5	0
		Expectations	/20	14	10
		Total	/70	3	0
ISYQOL/100	High	Spine-health	/100	2	0
TAASQ/5	High	Breast	/5	0	39
		Breast location	/5	0	75
		Breast shape	/5	0	47
		Breast size	/5	0	59
		Appearance	/5	0	8
		Clothing	/5	0	15
		Clothing general	/5	0	20
		Clothing specific	/5	0	25
BIDQ-S/5	Low	Total	/5	21	1

n: Number of participants, **ISYQOL:** Italian Spine Youth Quality of Life**, SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ v1.1:** Spinal Appearance Questionnaire version v1.1**, TAASQ:** Truncal Anterior Asymmetry Scoliosis Questionnaire**, BIDQ-S:** Body Image Disturbance Questionnaire-Scoliosis version.**, QOL:** Quality of life

5 CHAPTER 5 – TEST-RETEST RELIABILITY, AND CONVERGENT VALIDITY OF THE ISYQOL, TAASQ AND BIDQ-S IN ADULTS WITH SCOLIOSIS

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Abstract

Introduction: Adult Scoliosis (AS) has a significant impact on quality of life (QOL) as it can result in pain, disability, neurological impairment, and cosmetic concerns. New quality of life questionnaires including the Italian Spine Youth Quality of Life (ISYQOL) questionnaire, Truncal Anterior Asymmetry Scoliosis Questionnaire (TAASQ), and Body Image Disturbance Questionnaire-Scoliosis version (BIDQ-S) have recently been developed. As they were originally designed for use in the adolescent population, there is a need to evaluate their measurement properties in adults.

Objective: This study aimed to determine the test-retest reliability and the convergent validity of the ISYQOL, TAASQ, and BIDQ-S questionnaires against radiographic measurements and established quality of life questionnaires (SRS-22r and SAQ v1.1) in AS.

Study Design: A metrological study was conducted using a cross-sectional design for assessing validity and a single-group, test-retest design for assessing reliability.

Sample: Twenty-four consecutive participants with AS aged ≥18 years old and treated non-operatively were recruited from the offices of three spine surgeons and one physical therapy clinic.

Outcome Measures: Two established questionnaires (Scoliosis Research Society-22 refined (SRS-22r) and the Spinal Appearance Questionnaire (SAQ v1.1)) and three new questionnaires (ISYQOL, TAASQ, and BIDQ-S), were collected in addition to frontal radiographic

measurements (maximum Cobb angle, coronal balance, and both thoracic and lumbar vertebral rotations).

Methods: Questionnaires were administered electronically twice with a one-week interval between.

Results: The mean age of the participants was 30.7±18.7 years. Their average maximum curve angle was 41°±20°. Test-retest reliability estimates ($ICC_{3,1}$) for the new questionnaires ranged from 0.70 to 0.93. Convergent validity was demonstrated between the ISYQOL and SRS-22r (r=0.60) and SAQ v1.1 (r=-0.58) Total scores. Correlations between all domains and subdomains of the TAASQ and the SAQ v1.1 Total score. Appearance domain, and Expectations domain were between r = -0.32 and -0.72. The TAASQ Appearance and Clothing domains and their subdomains also correlated with the SRS-22r Self-image and Total scores (r = 0.43 to 0.75). Convergent validity of the BIDQ-S was supported by a high correlation with corresponding domain scores of the SRS-22r (Self-image domain: r = -0.82 and Total score: r = -0.72) and the SAQ v1.1 (Appearance domain: r = 0.43 and Total score: r = 0.44).

All domains and subdomains of the TAASQ demonstrated statistically significant correlations with the maximum Cobb angle except for the Appearance domain (r = -0.32 to -0.51). Only the TAASQ Breast domain and subdomains correlated with maximum thoracic rotation (r = -0.52 to -0.60). All TAASQ domains and subdomains except Breast and the Breast location domains demonstrated statistically significant correlation with Coronal balance (r = -0.35 to -0.53). The BIDQ-S only had a statistically significant correlation with Coronal balance (r=0.47), and the ISYQOL did not show

convergence with any radiograph measurements. All questionnaires except the ISYQOL and the BIDQ-S presented ceiling or floor effects (≥15%).

Conclusion: All new questionnaires demonstrated acceptable convergence with the established questionnaires, all except the ISYQOL showed convergence with radiograph measurements and all met the acceptable reliability threshold for research use. The TAASQ was the only questionnaire that met the acceptable threshold for both research and clinical use. The ISYQOL had the smallest MDC and SEM and was the only tool that did not present ceiling and floor effects.

5.1 Introduction

Adult Scoliosis (AS) is a spinal deformity in skeletally mature patients with Cobb angles of more than 10 degrees in the coronal plane.[98] AS consists of two major types: Adult Degenerative (or de novo) Scoliosis (ADS) and Adult Idiopathic Scoliosis.[12] The prevalence of ADS is as high as 68% in the healthy adult population over the age of 60, with an average age of 70.5 years. Conversely, Adult Idiopathic Scoliosis has a lower prevalence rate ranging from 0.4–3.9%.[9,10,22] AS has a significant impact on quality of life (QOL) as it may result in pain, disability, neurological impairment, and cosmetic concerns.[24]

Health-related QOL is a patient's assessment of the effects of a disease or treatment on their physical, psychological, and social domains of functioning and well-being.[55] Outcome measures assessing *health-related QOL* in AS are lacking. The Scoliosis Research Society (SRS-22) is the most common questionnaire used to measure QOL in adults with scoliosis.[53] However, it was not developed with patient input and was initially developed to measure QOL in candidates for surgery (i.e. severe scoliosis).[138 139,132] In a study of 173 adolescent females with IS who had undergone conservative treatment, nine items of the SRS-22 had major (≥50%) ceiling effects (participants with the best possible score), and 11 had moderate ceiling (≥20%) effects.[139] Such score distributions may limit the ability of the SRS-22r to detect positive response to conservative treatment.

The Spinal Appearance Questionnaire (SAQ v1.1) assesses the patient's perception of their appearance as viewed from the back, which may be less important to the patient compared to their

appearance viewed from the front.[85] Our findings in adolescents (refer to Chapter 3) showed that the SAQ v1.1 Total score had adequate reliability ($ICC_{3.1}$ = 0.86 to 0.94) and convergent validity with the SRS-22r Total score (r = -0.50). However, the SAQ v1.1 was also not developed with patients' input, which may affect its content validity, because it may be missing items that are of more importance to patients.[85] Also, the nine domain scoring strategy initially proposed for the SAQ is unlikely to detect subtle changes as each domain has a limited number of items.[85] Furthermore, the SAQ v1.1 was also not developed with Rasch scaling. [81] Rasch scaling would ensure that the score for each item will be given proper weight and reflect the severity of impact for the condition using the construct of interest.[132] Measurements developed with Rasch analysis have the attributes of a continuous measurement, theoretically providing more power in analyses and be better able to detect change.[132]

New questionnaires have been developed to address these limitations such as the Italian Spine Youth Quality of Life (ISYQOL), the Body Image Disturbance Questionnaire-Scoliosis version (BIDQ-S), and the Truncal Anterior Asymmetry Scoliosis Questionnaire (TAASQ). The ISYQOL questionnaire has recently been developed using Rasch analysis.[144] The ISYQOL items were developed with input from patients, families, and clinicians on an online forum to measure symptoms in patients with varying levels of disease and treatment (ranging from conservative treatment to surgical management).[81] This input is important to ensure that the questionnaire is comprehensive and related to real patient concerns.[81] Afterwards, a content analysis was performed using the concerns made in the online forum, generating a pool of 147 items. The relevance of these items was rated only by clinicians with expertise in spinal deformities.[81] The

ISYQOL has been translated into English by a consensus from four scoliosis experts which was subsequently reviewed by three Italian collaborators for compatibility with the original version.[83] The internal consistency of the ISYQOL has been tested in adolescents with idiopathic scoliosis and meets recommended internal consistency standards (α = 0.79 – 0.84).[83] However, measurements properties such as reliability and validity for the ISYQOL have not been studied in adults.

The Body Image Disturbance Questionnaire-Scoliosis version (BIDQ-S) is short, 7-question survey that includes appraisal of anterior appearance.[89] The Truncal Anterior Asymmetry Scoliosis Questionnaire (TAASQ) was also recently developed to evaluate self-perceived concerns from patients with AIS related to their anterior trunk appearance and the effects of these concerns on patient mindset and behavior.[140] However, measurement properties have not been established for the TAASQ in adults and adolescents with scoliosis.

Review of the current literature shows a lack of QOL outcome measures in the adult scoliosis population that are able to capture the concerns of patients with scoliosis. The limited availability of such measures may explain the lack of studies assessing the effects of conservative treatments for adults with scoliosis, as no existing outcome measures are sufficient to capture treatment-related changes.[145] Therefore, there is a need to assess if the measurements properties of the new outcome measures meet the minimum standards proposed by COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) for use in research and clinical settings.

5.2 Objective

The objective of this study is to determine the test-retest reliability, convergent validity, and ceiling and floor effects of the ISYQOL, TAASQ, and BIDQ-S questionnaires against established quality of life questionnaires (SRS-22r and SAQ V1.1) as well as the radiograph measurements in adults with scoliosis.

We hypothesized that:

(1) The test-retest reliability of the new questionnaires will be acceptable ($ICC_{3.1}$=0.70 – 0.90) and meet the minimum acceptable standard suggested by COSMIN for instruments used in measurements for groups of patients (i.e. research use, ICC ≥0.7) and in use for individual patients (i.e. clinical use, ICC ≥0.90).[132]

(2) The ISYQOL scores indicating better QOL will have moderate to good correlation (r ≥0.35) with scores indicating better QOL on the SRS-22r, better self-image on the SAQ v1.1, and with smaller radiographic measurements.[133]

(3) Scores on the TAASQ and BIDQ-S indicating better self-image would have moderate to good correlation (r ≥0.35) with scores indicating better QOL on the SRS-22r, better perceived appearance on the SAQ V1.1, and with smaller radiograph measurements.

(4) All questionnaires will be free from ceiling and floor effect or that such effects will be present at less than 15%.[132]

These hypotheses were based on the assumption that a larger the Cobb angle has been associated with scores reflective of poorer quality of life.[94,95] We expected stronger correlations among the

tools most focused on self-image/perceived appearance (SAQ V1.1, BIDQ-S, and TAASQ) and among the tools most focused on QOL in general (SRS-22r and ISYQOL). Additionally, higher correlations were expected between the tools most focused on self-image/perceived appearance (BIDQ-S, TAASQ) and the SAQ V1.1, or the SRS-22r self-image domain.

5.3 Methods

Study Design: A cross-sectional design was used for the convergent validity study and to determine ceiling and floor effects. A single-group, twice repeated-measures design was used for the test-retest reliability study. The study was approved by the University of Alberta Health Research Ethics Board (Pro00073569). (**Appendix 1)**

Participants: New participants were invited by the registration clerk at their consultation site (the Orthopedic Surgery Consultation Office and Curvy Spine Clinic in Alberta, Canada). Potential candidates who had discussed the study with the registration clerk were then asked to sign informed consent documents and complete the questionnaires prior to meeting their specialist. Originally, this study had a goal of enrolling 100 participants, however, recruitment was discontinued due to the COVID-19 pandemic. Therefore, analysis was only performed for the 24 participants enrolled in the study prior to closing recruitment. *Inclusion criteria* included the following: having a diagnosis of idiopathic scoliosis or degenerative (de novo) scoliosis, being >18 years old, having curve severity over 10°, and being fluent in English. *Exclusion criteria* included the following: having a history of spine surgery or other diseases that may affect quality of life and torso appearance, having a history of surgery or trauma in the torso or lower extremity,

and presenting with a positive neurological examination related to conditions other than neurological claudication or radiculopathy.

Procedure

Questionnaire data was obtained using the Research Electronic Data Capture (REDCap) server (version 7.6.9). These questionnaires were completed prior to the consultation with the scoliosis specialist. Questionnaires were collected on the same day as consultation and then collected again within 1 week following the collection of radiographs. Participants required roughly 15 minutes to complete all five questionnaires which were presented in the following order: ISYQOL, SRS-22r, SAQ v1.1, BIDQ-S, and TAASQ.

Radiograph Measurements

Prior to patient consultation with their specialist, radiographs were taken in standing position either as full-spine posterior-anterior radiographs using the EOS system at the University of Alberta Hospital or as standard digital full spine radiographs in other community clinics. Radiograph files were uploaded to the custom Medical Image Analysis Software (MIAS) Professional Edition (version 9.6.7.0) after being converted from Digital Imaging and Communications in Medicine (DICOM) formatting to a Joint Photographic Experts Group (JPEG) formatting. Maximal resolution for these images were maintained using ImageJ software (version 1.50i). (Rasband, W.S., ImageJ, U. S. National Institutes of Health, Bethesda, Maryland, USA, https://imagej.nih.gov/ij/, 1997-2018).

The maximum Cobb angle was subsequently measured by one rater (MA) using the MIAS software. Prior to data collection, MA, a physical therapist, was trained by observing one demonstration, discussing ten practice cases, and conducting these measurements twice ($ICC_{3,1}$= 0.99, 95% CI .94 to 1). The Cobb angle is a widely used measurement to evaluate curve severity and risk for progression, as well as help direct brace and surgical treatment in patients with scoliosis.[19] It is considered the most accurate and reliable method to quantify scoliosis severity, and a definitive diagnosis of scoliosis cannot be made without it.[15,19]

5.3.1 Cobb Angle:

To obtain the Cobb angle, a straight line was drawn along the upper endplate of the most tilted vertebra above the apex and along the lower endplate of the most tilted vertebra below the apex of each curve. The apex was identified by choosing the most translated vertebra in each of the spine curvatures. The angle between these two lines forms the Cobb angle.[24] All curves were measured and entered in the data file. The evaluator was blinded to the identity of the participants and the questionnaire scores during image measurement.

5.3.2 Axial vertebral rotation (AVR):

Stokes' method was used to measure AVR because it accounts for the three-dimensionality of the vertebra and is simple to use.[136] Stokes' method aims at measuring the axial rotation of vertebrae in degrees by measuring the projected distances of the center of each pedicle from the center of the vertebra, while accounting for the vertebra width and level.[136] The evaluator marked the position of the pedicle by positioning and adjusting the dimensions of ovals, then marked the

position of the lateral vertebral wall limit at the narrowest point, specifying the level before MIAS (the custom MIAS software (version 9.6.7.0) produced the rotation measurement.[136]

5.3.3 Coronal Balance

Coronal balance was measured as the horizontal distance between a vertical line drawn from the center of C7 and a second vertical line drawn from the center of S1.[137]

Measures of Quality of Life

5.3.4 Scoliosis Research Society-22 refined questionnaire (SRS-22r)

The Scoliosis Research Society-22 refined questionnaire (SRS-22r) consists of 22 items covering five domains: Pain, Self-Image, Function, Mental Health (all with five items each), and Satisfaction with Management (with only two items).[75] Scores ranging from 1 to 5 for each item are averaged to produce each domain score and a total score.[75] Higher scores on the SRS-22r indicate better quality of life. The original version of the SRS-22r (SRS-22), in a sample of 58 adolescents with AIS treated surgically, demonstrated acceptable reliability ($ICC_{3.1}$ = 0.96 to 0.85), concurrent validity ($r > 0.70$), and a 56.9% ceiling effect and 1.7% floor effect.[75] In a study including 73 adults and adolescents with idiopathic scoliosis, the SRS-22 was refined to improve the internal consistency of the Function domain which increased from $\alpha = 0.67$ to 0.78 leading to the current SRS-22r version.[134]

5.3.5 Spinal Appearance Questionnaire version 1.1 (SAQ v1.1)

The Spinal Appearance Questionnaire (SAQ v1.1) measures patient perceptions of their appearance related to spinal deformity.[88] The SAQ v1.1 consists of 14 items (including eight pictograms) regarding patient perceptions and expectations related to their appearance, with higher scores indicating poorer perceptions of symptoms resulting from scoliosis.[88] Each item of the SAQ v1.1 is scored from 1 to 5. The most current version of the SAQ (v1.1) was developed by Carreon et al.[88] Using factor analysis, they found that these 14 items loaded on two factors.[88] They proposed to sum the first 10 items as they related to the Appearance (SAQ Appearance domain) and the last 4 items as they related to Expectations (SAQ Expectations domain). The best possible total score for the SAQ is 14 and the worst possible score is 70. In the SAQ v1.1, patients must also identify which two aspects of their deformity that they find most bothersome. The last item is an open-ended question that asks patients which aspects of their appearance they wish to change, and why[11.] The study by Carreon et al. included 1,802 adolescents with AIS and demonstrated evidence for good reliability (Cronbach's $\alpha \geq 0.88$; test-retest correlation ≥ 0.81) and demonstrated evidence for convergent validity related to the major curve magnitude ($r = 0.32 – 0.36$).[88] In the adult population, the SAQ v1.1 correlated with the Self-image domain and Total score of the SRS-22r ($r = -0.53$ and $r = -0.60$, respectively), supporting convergent validity.[77]

5.3.6 Italian Spine Youth Quality Of Life Questionnaire (ISYQOL)

The ISYQOL is a new patient reported outcome measurement (PROM) for health-related quality of life that was proposed initially for use in adolescents with idiopathic scoliosis.[12] The ISYQOL

aims to measure symptoms in patients with varying levels of disease following treatments ranging from conservative treatment to surgical management.[12] Items of the ISYQOL were generated following a content analysis of the concerns expressed by patients, their parents, and scoliosis specialists in an online forum.[12] The ISYQOL was developed using Rasch analysis. Studies assessing the measurement properties of the ISYQOL in adolescents were promising. In a sample of 642 AIS adolescents (541 of whom were wearing braces as treatment), Caronni et al. concluded that the ISYQOL has better known-group validity and is better able to detect the impact of disease severity on health-related QOL in patients with AIS compared to the SRS-22.[142] To our knowledge, no measurement studies have been conducted in adults with scoliosis except for internal consistency which was reported as adequate by Zaina et al.[82]

5.3.7 Truncal Anterior Asymmetry Questionnaire (TAASQ)

The TAASQ was recently proposed to assess perceived anterior truncal appearance, as this measurement was missing from existing tools.[140] It consists of 14 items that fall into three main domains: Breast (questions 4 a, b, c; 12; 13; 14), Appearance (5; 6; 7; 10; 11), and Clothing (1; 2 a, b, c, d, e; 3; 8 a, b, c; 9). Two Clothing subdomains are further categorized as: Clothing-General (Q1, 3, 8a, 9) and Clothing-Specific (Q2, 8b, 8c, 8d). Three Breast subdomains are as follows: Breast Location (Q4c, 13), Breast Shape (Q4b, 12), and Breast Size (Q4a, 14). The TAASQ uses a combination of Likert scales and free text responses, making up 22 scored items. Each question provides a multiple-choice response featuring a scale ranging from 1 to 5, where 5 corresponds to the least amount of concern or perceived asymmetry (5 = best), and 1 corresponds to the most concern or highest amount of perceived asymmetry (1 = worst). The average score is then

calculated for each domain and sub-domain. Higher scores on the TAASQ indicate better quality of life.[140] To our knowledge, no studies have been published to investigate the measurement properties of the TAASQ in both adults and adolescents with scoliosis.

5.3.8 Body Image Disturbance Questionnaire - Scoliosis version (BIDQ-S)

The Body Image Disturbance Questionnaire - Scoliosis version (BIDQ-S) was derived from the generic BIDQ and adapted for use in assessing patients with scoliosis.[141] The BIDQ-S evaluates issues relevant to body image disturbance and is used to identify patients who are experiencing distress and impairment related to their appearance concerns.[141] The BIDQ-S consists of 7 items. Each answer is scored from 1 to 5, and the total score is obtained by calculating the mean of the seven items. Lower scores on the BIDQ-S indicate better QOL. The BIDQ-S (English version) demonstrated good internal consistency in adolescents with idiopathic scoliosis requiring surgery (Cronbach alpha = 0.82). It also showed convergent validity when compared to the SRS-22 Total score (r = -0.50 to -0.72).[141] A study including 259 adults and adolescents with idiopathic scoliosis demonstrated that the German version of the BIDQ-S has acceptable test-retest reliability ($ICC_{3.1}$= 0.79) and convergent validity with SRS-22r Total score (r= -0.72).[146] The treatments received by participants in this study were not specified.

5.4 Statistical Analysis

5.4.1 Sample Description

Demographic data (age and sex) were summarized, and the outliers were reviewed using explore analysis in SPSS version 24.0 (SPSS, Inc., Chicago, IL, USA). For continuous data, the average, range, and standard deviation are presented.

The following statistical analyses were performed:

- The Intra-class Correlation Coefficient ($ICC_{3,1}$) was used to examine the test-retest reliability between surveys collected at the first visit and one-week following at the second visit. The Minimal Detectable Change 95 (MDC 95) and the Standard Error of Measurement (SEM) was reported.[78]
- Pearson product-moment correlation coefficient analysis was used to evaluate the correlations between the questionnaires scores and both established questionnaires scores and radiographic measurements. One-tailed significance levels were used for the directional hypothesis testing.
- Ceiling and floor effects were determined by calculating the percentage of participants scoring at the best and worst possible score, respectively.[132]

5.5 Results

Participants included 24 adults with scoliosis (17 females and 7 males) with a mean age of 30.7±18.7 years and a maximum curve angle of 41.0° ±20.0°. Of the participants, 14 received conservative treatment (3 had a brace, 11 received exercise treatment, and the others were under

observation). Mean total QOL scores on the SRS-22r and SAQ v1.1 were 3.3±0.7 and 40.4±13.1, respectively **(Table 5-1)**. Characteristics of the 15 participants in the reliability study are reported in **Table 5-1.** All characteristics were similar to those of the validity sample, but their mean curve angle was slightly larger (47.0 °±18.1° vs. 41.0 °±20.0 °).

5.5.1 Test-retest reliability

The test-retest reliability of the ISYQOL was $ICC_{3,1}$ =0.70. The test-retest $ICC_{3,1}$ for the BIDQ-S was 0.77.(**Table 5-3**) All the TAASQ domain scores had test-retest $ICC_{3,1}$ of 0.74 or larger. These values for $ICC_{3,1}$ would be 0.83 or larger if subdomains were excluded. The Total scores of the SAQ v1.1 and the SRS-22r both demonstrated a high test-retest reliability ($ICC_{3,1}$= 0.90). The test-retest reliability was 0.79 or larger ($ICC_{3,1}$) for the SRS-22r domain scores, and 0.73 or larger for the SAQ v1.1 domains.

The percentage of scales corresponding to the SEM and MDC for the newer questionnaires ranged between 6.2% to 10.2% and 17.4% to 28.6%, respectively (**Table 5-3).** For the SRS22r, the SEM varied between 3.6% and 7.6% of the scale while that of the SAQ v1.1 varied from 6.0% to 26.4%. The TAASQ Clothing general domain had the highest SEM and MDC among the new questionnaires (10.2% and 28.6%, respectively), whereas the lowest SEM and MDC were observed for the TAASQ Clothing specific domain (6.2% and 17.4%, respectively) and the ISYQOL (6.8% and 18.7%, respectively).

5.5.2 Convergent Validity with Questionnaires

The ISYQOL score correlated significantly with all the SRS-22r scores (r = 0.4 to 0.6, respectively) and with all the SAQ v1.1 scores (r = -0.42 to -0.58). The TAASQ Appearance, and Clothing main domain scores correlated significantly with the SRS-22r Total score (r= 0.53 and 0.50, respectively) and with the SRS-22r Self-Image and Mental Health domains (r = 0.5 to 0.75). The TAASQ Breast score did not correlate significantly with the SRS-22r scores, and the TAASQ did not correlate significantly with the SRS-22r Pain and Function domains. Among the TAASQ subdomains, only the Clothing general and Specific sub-scores demonstrated significant correlations (r=0.43 to 0.66) with the Total, Self-Image, and Mental Health scores of the SRS-22r. All the TAASQ domain and subdomain scores significantly correlated with the SAQ v1.1 scores (r= -0.40 to -0.72) with the exception of the Breast location subdomain (**Table 5-4**). The BIDQ-S correlated significantly with all the SRS-22r scores (r= -0.46 to -0.82). The BIDQ-S also correlated significantly with the SAQ v1.1 Total (r=0.44) and Appearance domain (r=0.43) scores (**Table 5-4**).

5.5.3 Convergent Validity with Radiograph Measurements:

The ISYQOL score did not correlate significantly with the radiographic measurements (r = -0.32 to 0.09). A significant correlation was found between the TAASQ Breast or the Clothing main domain (but not with Appearance) and maximum Cobb angle (r = -0.48, -0.41 and -0.32, respectively). Only the TAASQ Breast domain and subdomains correlated significantly with maximum thoracic rotation (r = -0.52 to -0.60) (**Table 5-5**). The TAASQ Appearance and Clothing

domains, the Clothing subdomains, and two of Breast subdomains (Breast shape and size) correlated significantly with coronal balance (r= -0.35 to -0.53). The only radiographic measurement that correlated significantly with the BIDQ-S was coronal balance (r= 0.47). None of the questionnaires correlated with lumbar rotation (**Table 5-5**).

5.5.4 Ceiling and Floor Effects

The ISYQOL showed no ceiling or floor effects. The Breast domains of the TAASQ presented the highest floor effects: Breast= 18%, Breast location= 31.2%, Breast shape = 27.3%, and Breast size = 22.7%. For the BIDQ-S, 4% of the participants scored the best (ceiling effect), and no participants scored at the worst possible score (floor effect). In contrast, the SRS-22r showed no floor effects, and the percentage of ceiling effects in the affected domains of the SRS-22r were as follows: Function= 12%, Pain= 5%, Mental Health= 4%, and Satisfaction= 8%. Ceiling effects for the SAQ v1.1 were: Appearance= 4% and Expectations= 4% while a floor effect was observed only in the Expectation domain (16%) (**Table 5-6**).

5.6 Discussion

The scores of all questionnaires (domains and subdomains) that have been examined in this study (SRS-22r, SAQv1.1, ISYQOL, TAASQ, and BIDQ-S) demonstrated adequate test-retest reliability for research use (ICC≥0.70). The SRS-22r (Function domain score, Pain domain score, and Total score), SAQ v1.1 (Appearance domain and Total score), and the TAASQ (Breast domain, Breast size subdomain, and the Appearance domain) also show adequate reliability for individual use (ICC≥0.90).

Overall, the ISYQOL, TAASQ (except for the Breast location domain), and BIDQ-S (except vs. the SAQ v.1.1 Expectations domain) demonstrated convergent validity by significantly correlating with the SAQ v1.1 (Table 5-4). Similarly, the ISYQOL, TAASQ (except the Breast domain), and BIDQ-S demonstrated convergent validity by significantly correlating with the SRS-22r (Table 5-4). Finally, while the TAASQ and BIDQS showed correlation with radiographic measurements, the ISYQOL did not (Table 5-5).

To our knowledge, no previous studies have examined the measurement properties of the BIDQ-S and TAASQ in adult patients with scoliosis. However, the BIDQ-S has previously demonstrated good internal consistency in adolescents with idiopathic scoliosis who required surgery (Cronbach alpha = 0.82). Convergent validity with the SRS-22 and Total score (r = -0.50 to -0.72) was also found.[141] Only internal consistency of the ISYQOL has been reported in adult patients. Zaina and colleagues evaluated the internal consistency of the ISYQOL in adults with idiopathic scoliosis and also found it to be acceptable (α= >0.7), suggesting that the ISYQOL could be a useful tool to measure quality of life in adults with scolisis.[82] However, direct comparison between findings from the current study and previous metrological studies in adults is not possible.

Interestingly, the Breast domain of the TAASQ demonstrated convergence only with the SAQ Appearance and Total scores. This may be attributable to pictograms provided in the SAQ, that may help the patient to precisely describe their cosmetic issues. Also, convergence with Cobb angle was found only with the more appearance-related TAASQ domains (Breast and Clothing) but excluded the Appearance domain. Overall, most of the TAASQ questions are clothing and breast related. This might explain the low convergence of the Appearance domain with the Cobb

angle. Furthermore, no significant correlations were found between the lumbar rotation and any of the new questionnaires. This may be attributed to the soft tissue surrounding the lumbar spine, which may deform and mask the deformity resulting from scoliosis. Among the participants, 22 presented with thoracic curves with average Cobb angles of $42^{o}\pm 20^{o}$ while only 19 presented with lumbar curves with average Cobb angles of $36^{o}\pm 15^{o}$. Thoracic vertebral rotation demonstrated convergence with only Breast domains and subdomains, which is logical given that thoracic deformity is more likely to affect breast appearance than any other changes reflected in our radiographic measurements. Additionally, coronal balance correlated significantly with the BIDQ-S and the following TAASQ scores: Breast shape, Breast size, Appearance, Clothing, Clothing General general, and Clothing Specific domains. Finally, no significant correlation was found between and the ISYQOL and the coronal balance. This can be explained by the nature of the ISYQOL which is a questionnaire assessing QOL, rather than self-image perceptions.

Despite the absence of evidence regarding the convergence for the ISYQOL with radiograph measurements, this study supports the careful use of the ISYQOL for QOL research in adult scoliosis. The ISYQOL had adequate convergence with the other QOL tools. The ISYQOL appears more likely to detect changes than the SRS-22r and the SAQ v1.1 since its score is free from ceiling and floor effects and its MDC95 was among the smallest of the possible scores. However, longitudinal responsiveness studies are needed before recommending wider implementation of the ISYQOL in order to both quantify the ability of the ISYQOL to detect changes over time and determine the magnitude of changes matter to patients.

The BIDQ-S and TAASQ demonstrated an acceptable level of measurement reliability and convergent validity, with the TAASQ demonstrating convergent validity with both established questionnaires and radiograph measurements. Additionally, the TAASQ presented higher reliability than established questionnaires and radiograph measurements. Both the BIDQ-S and TAASQ captured anterior perceived appearance which is hypothesized to have more relevance to patients than the perceived posterior appearance assessed by the SAQ v1.1. However, the TAASQ and the SAQ v1.1 still contain ceiling or floor effects. The BIDQ-S is shorter than both the SAQ v1.1 and the TAASQ; it has less ceiling and floor effects and presented the smallest SEM and MDC compared to the SAQ v1.1 and the TAASQ.

Based on our results, we can support the use of all the new questionnaires (ISYQOL, TAASQ, and BIDQ-S) in research settings. Additionally, the TAASQ Appearance and Breast domains met COSMIN criteria for individual, clinical use. However, an outcome measure administrator should be aware of the following factors when selecting which tool to use. First, consider the limitations of the questionnaires. For instance, one should be careful when interpreting the scores of the questionnaires as they may present with ceiling or floor effects. Clustering of patient scores at the extremes of the questionnaire scales affects that questionnaire's ability to detect further improvement or deterioration, rendering it useless in assessing further change for a patient as it relates to treatment outcomes.

Secondly, one should consider the context in which the measurement will be used. For example, in situations where time is limited, one might choose the BIDQ-S over the other questionnaires as it is the shortest questionnaire. Lastly, one must take into consideration the decisions that will be

made based on the results. For example, measurements used to discriminate between patients needing major surgery and those who don't, should have higher convergent validity in relation to radiographic measurements as more serious clinical consequences hinge on the measurement results. In contrast, a questionnaire measuring improvements in perceived appearance or quality of life to document progress in rehabilitation may be adequate with measurement properties meeting minimum reliability and validity standards if it can better detect changes.

5.6.1 Strengths and Limitations

This novel study examined measurement properties of the ISYQOL, TAASQ, and BIDQ-S in adults with scoliosis. Its limitations include limited generalizability due to small sample size and recruitment of participants from a single, specialized clinic. Additionally, this study presents with limited generalizability as it excluded surgical candidates. For this study, participants were limited to patients being treated conservatively as this is an understudied population. Additionally, previous studies have stated concerns that existing assessments may not perform well in this population. Originally, this study aimed at recruiting 100 participants. However, due to the COVID-19 pandemic, the recruitment process was ended prior to reaching recruitment goals. Lastly, sagittal views of the spine were not available for many patients. Therefore, convergence between questionnaires and sagittal balance, which has been reported as the most important and reliable radiographic predictor of clinical health status in adults with scoliosis, could not be assessed.[147]

5.6.2 Future Directions

We suggest that future studies should evaluate the responsiveness of these questionnaires and determine to what extent these questionnaires are able to detect change. Furthermore, this study featured the use of Classical Test Theory (CTT); however, future researchers should consider the use of generalizability theory in order to identify the most important sources of score variation affecting repeatability.[132] Additionally, future studies should evaluate how these questionnaires perform in different languages, with different age groups, and with other spinal deformities. Another direction for future studies could include the development of new questionnaires for adults with scoliosis, ensuring that items are relevant and comprehensive to adults because all questionnaires examined in this study were originally developed to assess QOL in adolescent patients. Relevance of survey items should be assessed for three aspects: (1) relevance to the construct, (2) relevance to population (i.e. age and sex), and (3) relevance to the purpose of the measurement (discriminative, evaluative, or predictive)[132,148]. Comprehensiveness can be assessed by asking the following question: "Is the construct completely covered by items?"[132]

5.6.3 Conclusion

All new questionnaires demonstrated acceptable convergent validity with established questionnaires as well as with the radiograph measurements (except the ISYQOL). All questionnaires met the reliability threshold for research use. The TAASQ was the only questionnaire meeting the reliability threshold for both research and individual, clinical use. The ISYQOL and the BIDQ-S had smaller MDC and SEM and The ISYQOL was the only tool free

from ceiling and floor effects. Overall, the TAASQ is valid, and sufficiently reliable for both research and clinical use, while the ISYQOL and the BIDQ-S are valid, and reliable for research use.

Table 5-1: Description of the Participants

	Validity sample			Reliability sample		
	n	Range	Mean (SD)	n	Range	Mean (SD)
Age (years)	24	18; 81	30.7 (18.7)	15	18; 64	31.1 (17.0)
Maximum Cobb Angle (°)	23*	10; 76	41.0 (20.0)	14*	10.5; 76	47.0 (18.1)
Maximum Thoracic Rotation (°) **	22	-9.5; 17.7	7.9 (6.9)	13	-9.5; 16.3	7.8 (6.4)
Maximum Lumbar Rotation (°) **	22	-21.5; -13.8	-5.4 (9.5)	13	-21.3; 11	-6.4 (9.3)
Coronal Balance (mm)	24	0.3; 52.1	10.1 (11.6)	15	0.3; 52.1	11.4 (13.9)

n: Number of participants
SD: Standard deviation
°: Degree
* **1** image not available
** not all participant had a thoracic and lumbar curve to measure

Table 5-2: Means and Standard Deviation for All Questionnaires at All Time Points.

Questionnaire	Good QOL	Domains		Mean (SD)	
				Baseline (n=24)	One week (n= 15)
SRS-22	High	Function	/5	3.9 (0.7)	3.9 (0.9)
		Pain	/5	3.3 (0.7)	3.5 (0.7)
		Self-image	/5	2.9 (0.8)	3.0 (0.8)
		Mental health	/5	3.5 (0.8)	3.5 (0.8)
		Satisfaction	/5	2.9 (1.0)	3.0 (0.9)
		Total	/5	3.3 (0.6)	3.4 (0.6)
SAQ v1.1	Low	Appearance	/50	26.3 (10.3)	26.4 (9.3)
		Expectations	/20	14.1 (4.3)	13.4 (4.9)
		Total	/70	40.4 (13.1)	39.8 (13.0)
ISYQOL	High	Spine health	/100	41.8 (9.8)	46.5 (6.2)
TAASQ	High	Breast	/5*	3.9 (0.9)	3.9 (1.1)
		Breast location	/5	4.2 (0.9)	4.1 (1.3)
		Breast shape	/5	3.9 (1.1)	3.9 (1.2)
		Breast size	/5	3.7 (1.2)	3.6 (1.2)
		Appearance	/5	3.3 (1.2)	3.4 (1.1)
		Clothing	/5	3.0 (0.8)	3.1 (1.0)
		Clothing general	/5	2.9 (1.0)	2.8 (1.0)
		Clothing specific	/5	3.2 (0.8)	3.4 (1.1)
BIDQ-S	Low	Total	/5	2.3 (1.1)	2.3 (0.8)

n: Number of participants**, ISYQOL:** Italian Spine Youth Quality of Life**, SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ V1.1:** Spinal Appearance Questionnaire**, TAASQ:** Truncal Anterior Asymmetry Scoliosis Questionnaire**, BIDQ-S:** Body Image Disturbance Questionnaire-Scoliosis version**, SD:** Standard deviation, **QOL:** Quality of life, *: n=14

Table 5-3: Test-retest Reliability of Questionnaire Scores

Tools	Good QOL	Domains		Test-retest Reliability pre-visit vs. one week			
				N	ICC $_{3,1}$ (95 CI)	SEM (% of score)	MDC95 (% of score)
SRS-22	High	Function	/5	15	0.90 (0.75 to 0.97)	0.2 (4.2)	0.5 (9.0)
		Pain	/5	15	0.95 (0.86 to 0.98)	0.2 (3.6)	0.7 (13.4)
		Self-image	/5	15	0.84 (0.60 to 0.94)	0.3 (6.2)	0.5 (10.2)
		Mental health	/5	15	0.79 (0.50 to 0.92)	0.4 (7.6)	0.9 (17)
		Satisfaction	/5	15	0.87 (0.67 to 0.95)	0.3 (6.8)	0.9 (18.8)
		Total	/5	15	0.90 (0.74 to 0.96)	0.2 (3.6)	0.5 (9.8)
SAQ v1.1	Low	Appearance	/50	15	0.92 (0.78 to 0.97)	1.3 (26.4)	3.7 (7.3)
		Expectations	/20	15	0.73 (0.38 to 0.90)	2.4 (12.2)	6.7 (33.7)
		Total	/70	15	0.90 (0.73 to 0.96)	4.2 (6.0)	11.7 (16.6)
ISYQOL	High	Spine health	/100	15	0.70 (0.33 to 0.88)	6.8 (6.8)	18.7 (18.7)
TAASQ	High	Breast	/5	14*	0.90 (0.73 to 0 .97)	0.3 (6.6)	0.9 (18.0)
		Breast location	/5	14*	0.86 (0.63 to 0.95)	0.3 (6.4)	0.9 (18.0)
		Breast shape	/5	14*	0.80 (0.50 to 0.93)	0.4 (7.4)	1.0 (20.6)
		Breast size	/5	14*	0.93 (0.81 to 0.98)	0.3 (6.4)	0.9 (17.8)
		Appearance	/5	15	0.92 (0.80 to 0.97)	0.5 (9.6)	1.3 (26.4)
		Clothing	/5	15	0.83 (0.59 to 0.94)	0.4 (8.0)	1.1 (22.4)
		Clothing general	/5	15	0.85 (0.85 to 0.93)	0.5 (10.2)	1.4 (28.6)
		Clothing specific	/5	15	0.74 (0.40 to 0.90)	0.3 (6.2)	0.9 (17.4)
BIDQ-S	Low	Total	/5	15	0.77 (0.46 to 0.91)	0.5 (9.2)	1.3 (25.8)

n: Number of participants**, ISYQOL:** Italian Spine Youth Quality of Life**, SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ V1.1:** Spinal Appearance Questionnaire**, TAASQ:** Truncal Anterior Asymmetry Scoliosis Questionnaire**, BIDQ-S:** Body Image Disturbance Questionnaire-Scoliosis version.**, ICC:** Intra-class Correlation Coefficient**, SEM:** Standard Error of Measurement**, MDC:** Minimal Detectable Change**, CI:** Confidence Interval**, QOL:** Quality of life ***: Item not answered due to irrelevance**

Table 5-4: Pearson Correlation Estimates for Convergent Validity Between Newer and Established Questionnaires Scores.

		SAQ v1.1			SRS-22r					n
		Appearance	**Expectations**	**Total**	**Function**	**Pain**	**Self-Image**	**Mental Health**	**Total**	
ISYQOL - Spinal Health	r	-.52**	-.42**	-.58**	.46**	.58**	.58**	.40**	.60**	24
TAASQ - Breast	r	-.61**	-.43**	-.63**	.04	-.07	.34	.20	.12	22
TAASQ - Breast location	r	-.54**	-.32	-.57**	.09	.09	.30	.23	.18	22
TAASQ - Breast shape	r	-.60**	-.43**	-.60**	.12	<-.01	.32	.33	.20	22
TAASQ - Breast size	r	-.53**	-.40*	-.54**	.04	-.11	.38	.18	.10	22
TAASQ - Appearance	r	-.56**	-.54**	-.63**	.20	.26	.75**	.60**	.53**	22
TAASQ - Clothing	r	-.63**	-.42*	-.65**	.34	.34	.63**	.50*	.50*	24
TAASQ - Clothing general	r	-.67**	-.40*	-.72**	.20	.26	.66**	.46*	.43*	24
TAASQ - Clothing Specific	r	-.62**	-.41*	-.63**	.38	.39	.56**	.45*	.52**	24
BIDQ-S Total	r	.43*	.39	.44*	-.46*	-.48*	-.82**	-.64**	-.72**	24

n: Number of participants, **r:** Pearson correlation coefficient, **ISYQOL:** Italian Spine Youth Quality of Life**, SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ v1.1:** Spinal Appearance Questionnaire version 1.1**, TAASQ:** Truncal Anterior Asymmetry Scoliosis Questionnaire**, BIDQ-S:** Body Image Disturbance Questionnaire-Scoliosis version**.**

** Correlation is significant at the 0.01 level (2-tailed). * Correlation is significant at the 0.05 level (2-tailed).

Table 5-5:Correlation Between Questionnaires Scores and Radiograph Measurements.

	Good QOL	Maximum Cobb Angle r (n)	Maximum Thoracic Rotation r (n)	Maximum Lumbar Rotation r (n)	Coronal Balance r (n)
ISYQOL - Spinal health -	High	-.32 (23)	-.24 (22)	.09 (22)	.07 (24)
TAASQ - Breast	High	-.48* (20)	-.60** (19)	-.12 (19)	-.32 (21)
TAASQ - Breast location		-.40* (20)	-.60** (19)	-.10 (19)	-.06 (21)
TAASQ - Breast shape		-.39* (20)	-.53** (19)	-.20 (19)	-.38* (21)
TAASQ - Breast size		-.51* (20)	-.52* (19)	-.05 (19)	-.42* (21)
TAASQ - Appearance		-.32 (22)	-.30 (21)	-.06 (21)	-.53** (23)
TAASQ - Clothing		-.41* (22)	-.26 (21)	-.04 (21)	-.41* (23)
TAASQ - Clothing general		-.44* (22)	-.23 (21)	-.03 (21)	-.41* (23)
TAASQ - Clothing specific		-.39* (22)	-.27 (21)	.00 (21)	-.35* (23)
BIDQ-S Total	Low	.12 (23)	.07 (22)	-.08 (22)	.47** (24)

r: Pearson correlation coefficient **n:** Number of participants**, ISYQOL:** Italian Spine Youth Quality of Life**, SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ V1.1:** Spinal Appearance Questionnaire version 1.1**, TAASQ:** Truncal Anterior Asymmetry Scoliosis Questionnaire**,** and **BIDQ-S:** Body Image Disturbance Questionnaire-Scoliosis version.

** Correlation is significant at the 0.01 level (2-tailed). * Correlation is significant at the 0.05 level (2-tailed).

Table 5-6: Percentage of the Sample with Ceiling and Floor Effects for Each of the Questionnaire Scores.

Questionnaires	Good QOL	Domains		Ceiling (%)	Floor (%)
SRS-22	High	Function	/5	12.0	0.0
		Pain	/5	5.0	0.0
		Self-image	/5	0.0	0.0
		Mental health	/5	4.0	0.0
		Satisfaction	/5	8.0	4.0
		Total	/5	0.0	0.0
SAQ V1.1	Low	Appearance	/50	4.0	0.0
		Expectations	/20	4.0	16.0
		Total	/70	0.0	0.0
ISYQOL	High	Spine health	/100	0.0	0.0
TAASQ	High	Breast	/5	0.0	18.0
		Breast location	/5	0.0	31.2
		Breast shape	/5	0.0	27.3
		Breast size	/5	4.5	22.7
		Appearance	/5	4.2	0.0
		Clothing	/5	0.0	0.0
		Clothing general	/5	0.0	4.2
		Clothing specific	/5	4.2	0.0
BIDQ-S	Low	Total	/5	4.0	0.0

n: Number of participants, **ISYQOL:** Italian Spine Youth Quality of Life**, SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ V1.1:** Spinal Appearance Questionnaire version 1.1**, TAASQ:** Truncal Anterior Asymmetry Scoliosis Questionnaire**, BIDQ-S:** Body Image Disturbance Questionnaire-Scoliosis version.**, QOL:** Quality of life

6 CHAPTER 6 – DISCUSSION

Scoliosis is "a complex three-dimensional rotational deformity that affects the spine in the coronal, sagittal, and axial planes" that significantly impacts Quality of Life (QOL) in adolescents and adults with scoliosis.[1] In order to improve QOL for patients with scoliosis and provide more options for symptom relief, understanding the effects of conservative treatments such as therapeutic exercise for both adolescents and adult patients with scoliosis is necessary. The effects of conservative treatments on adolescents with scoliosis has been documented in previous systematic reviews.[149–151] However, following a systematic review (refer to Chapter 2), we found only one study meeting our selection criteria, suggesting there is a need for more studies examining the effects of conservative treatment on adults with scoliosis.[4,5] Additionally, the study meeting our selection criteria was found to present a high risk of bias due to the lack of blinding for both participants and assessors. Given these observations, there is a need for high quality studies examining the effect of exercise on QOL in adults with scoliosis. However, to conduct such studies, there is both need for QOL outcome measurements with adequate measurement properties to capture changes following treatment and studies that compare these assessments.

Several patient-reported outcome measures (PROMs) assessing QOL in patients with scoliosis are available. Currently, both the SRS-22r and SAQ are commonly used to assess QOL in patients with scoliosis. However, they suffer from having poor measurement properties, thereby limiting proper measurement of QOL.[152,77] Therefore, new PROMs, namely the ISYQOL, TAASQ, and BIDQ-S, used for QOL assessment have been developed to address the limitations of the SRS-22r and SAQ. However, despite the development of new PROMs, there is a lack of published studies

examining the measurement properties of these PROMs in comparison to the older PROMs in adolescents and adults with scoliosis. Comparison of these PROMs were needed in order to determine their ability to adequately evaluate the efficacy of treatment options.

This thesis, therefore, aimed to compare the measurement properties of newly developed questionnaires in both adolescents and adults with scoliosis (refer to Chapter 4 and 5). Initially, our thesis aimed to use the SRS-22r and the original SAQ questionnaire[85] to compare to reliability and validity of the new questionnaires. Recently, however, a new version of the SAQ, the SAQ v1.1, was developed. Therefore, this thesis also sought to compare the original SAQ and the SAQ v1.1 to decide which one had better measurement properties for use as a reference when evaluating the new tools (refer to Chapter 3). Three metrological studies were conducted in this thesis in accordance with the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN).[132] In this chapter, results of the previous chapters will be summarized and then discussed in terms of applications, limitations, and future directions. Additionally, the discussion will include suggestions to inform questionnaire selection based on different clinical situations.

6.1 Summary of Results

6.1.1 Measurement Properties of the SAQ and SAQ v1.1 Questionnaires in Adolescents with Scoliosis

The test-retest reliability of the SAQ domains ($ICC_{3,1}$ from 0.72 to 0.94) in adolescents with scoliosis was similar or slightly lower than that for the scores of the SAQ v1.1 Total score,

Appearance domain, and Expectations domain $ICC_{3,1}$ (95%CI) = 0.92 (0.87 – 0.96), 0.94 (0.89 - 0.97), and 0.86 (0.75 - 0.92), respectively.

Additionally, we found that the correlations between SAQ scores and both the SRS-22r Total score (r= -0.35 to -0.59) and Cobb angle (r= 0.38 to 0.59) provided evidence for convergent construct validity in all domains of the SAQ except for the Curve and Chest domains. However, the evidence for convergent validity was stronger for the SAQ v1.1, compared to the SAQ, as shown by the correlation between the SAQ v1.1 Total score with the SRS-22r Total score (r=-0.50) and Cobb angle (r=0.56). Similarly, the Total score of the SAQ v1.1 demonstrated convergent validity with thoracic rotation (r = 0.40). Correlation of the SAQ v1.1 and other radiograph measurements ranged from r = 0.12 to 0.46. A low and non-significant correlation was observed between all domains of both SAQ versions and lumbar rotation except for the SAQ Curve domain (r = 0.37). However, we hypothesized that this finding was due to the fact that lumbar deformity is often masked by overlaying soft tissue, possibly reducing the impact of lumbar rotation on negatively perceived appearance.

Results from ceiling and floor effect analyses also favored the newer SAQ v.1.1. Such effects were only observed for the following domains of the original SAQ: Curve= 11% (ceiling effect), Kyphosis= 68% (ceiling effect), and Waist= 4% (floor effect). For the SAQ v1.1, ceiling effects were observed for the following: Total score = 3%, Appearance = 5%, and Expectation = 14%. Floor effects for the SAQ v1.1 were observed only for the Expectations domain (10%). These findings suggested that the SAQ v1.1 is a better instrument at assessing change in patients with

scoliosis compared to the SAQ. Therefore, the SAQ v1.1 was used for the subsequent studies included in this thesis as a reference test along with the SRS-22r and radiographic measures.

6.1.2 Measurement Properties of New Questionnaires in Adolescents with Scoliosis

In Chapter 4, we found that the test-retest reliability estimate ($ICC_{3,1}$) for the new questionnaires in adolescents ranged from 0.77 to 0.95. Convergence was demonstrated between the ISYQOL and both the SRS-22r (r=0.67) and the SAQ v1.1 Total scores (r= -0.53). Additionally, a significant correlation was found between the TAASQ Breast, Appearance, and Clothing scores and the SAQ v1.1 Total score, Appearance domain, and Expectations domain; these correlations ranged between r = -0.44 and -0.69 and demonstrate convergent validity between the TAASQ and the SAQ v1.1. Similarly, the main domains of the TAASQ correlated with the SRS-22r Self-image and Total scores (r= 0.42 to 0.70) supporting convergent validity between these measures. The convergent validity of the BIDQ-S was supported by a high correlation to both the SRS-22r Total score and the SAQ v1.1 Total score (r= -0.65 and 0.62, respectively). Additionally, the BIDQ-S demonstrated evidence of convergent validity with appearance domains from established questionnaires (SRS-22r Self-image r= -0.55 and SAQ v1.1 Appearance r=0.60).

The ISYQOL did not demonstrate convergent validity with radiographic measurements. Significant correlations were found between the main domains of the TAASQ (Breast, Appearance, and Clothing) and maximum Cobb angle, thoracic vertebral rotation, and coronal balance (r = -0.24 to -0.47), except for a low correlation between the Clothing domain and coronal balance where r = -0.13. The BIDQ-S showed a significant correlation with the maximum Cobb

angle, thoracic vertebral rotation, and coronal balance (r = 0.35, 0.33, and 0.23, respectively). As observed in the SAQ v1.1 comparison study, none of the new questionnaires correlated with lumbar rotation.

Only the ISYQOL did not present notable ceiling or floor effects (≥15%) as only 2% of respondents presented ceiling effects and none had floor effects. Conversely, the SRS-22r, SAQ v1.1, TAASQ, and BIDQ-S demonstrated ceiling effects between 2% and 22% and floor effects between 1% and 75%. These findings, according to COSMIN, suggest that the TAASQ Appearance and Clothing domains are valid and reliable for both research and clinical use, whereas the Breast domain of the TAASQ, the ISYQOL, and the BIDQ-S are valid and reliable for research use only. Overall, the analysis of ceiling effects favours the use of the ISYQOL.

6.1.3 Measurement Properties of New Questionnaires in Adults with Scoliosis

In Chapter 5, the measurement properties for the new questionnaires was evaluated in adults with scoliosis. Results of that study showed that the test-retest reliability estimate ($ICC_{3,1}$) for the new questionnaires ranged from 0.70 to 0.93. Adequate convergent validity (r≥0.35) was demonstrated between the ISYQOL and all of the scores from the SRS-22r and SAQ v1.1. Significant correlations between the TAASQ Breast, Appearance, and Clothing scores and the SAQ v1.1 Total score, Appearance domain, and Expectations domain ranged between r = -0.42 and -0.65. The TAASQ Appearance and the Clothing domains and their subdomains also showed adequate correlations (r≥0.35) with the SRS-22r Self-image score and Total score (r = 0.63 to 0.75). The

adequate convergence (r≥0.35) of the BIDQ-S was supported by a high correlation with all SRS-22r scores and with the SAQ v1.1 Total score and Appearance score.

The ISYQOL score did not correlate significantly with radiographic measurements (r = -0.32 to 0.09). A significant correlation was found between the TAASQ Breast and Clothing main domain and the maximum Cobb angle (r = -0.48 and -0.41, respectively), but significant correlation was not found for the TAASQ Appearance score. Only the TAASQ Breast domain and subdomains correlated significantly with maximum thoracic rotation (r = -0.52 to -0.60) **Table 5-5**. The TAASQ Appearance and Clothing domains, including the Clothing subdomain and the two subdomains for Breast (Breast Shape and Size), correlated significantly with coronal balance (r= -0.35 to -0.53). The only radiographic measurement that correlated significantly with the BIDQ-S in adults with scoliosis was coronal balance (r= 0.47). As in previous chapters, none of the questionnaires correlated with lumbar rotation.

All questionnaires presented some ceiling or floor effects except the ISYQOL (0%). However, in adults with scoliosis, no questionnaire presented ceiling effects exceeding 15%, and only the SAQ v1.1 Expectations, and the TAASQ Breast scores presented floor effects >15%. The SRS-22r, SAQ v1.1, non-Breast TAASQ domains and subdomains, and BIDQ-S demonstrated ceiling effects ranging between 0% and 12% and floor effects ranging between 0% and 4.2%. Overall, the TAASQ Appearance, Breast domain, and Breast size subdomain are valid and reliable for both research and clinical use according to COSMIN. However, the TAASQ Breast shape subdomain, Breast location subdomain, and Clothing domains, along with the ISYQOL and the BIDQ-S are

valid and reliable for research use only. All of the new questionnaires except for the TAASQ breast-related scores presented adequate ceiling and floor effect levels.

6.2 Interpretation of the Findings from Each Chapter

Although findings from each study have been discussed in previous chapters, in this chapter we will briefly discuss the following: 1) the findings of each study, 2) comparison between the measurement properties of the new questionnaires to older assessments in adolescents and adults with scoliosis, 3) the strengths and limitations of these studies, and 4) suggestions to inform on implications and future directions.

6.2.1 Measurement Properties of the SAQ and SAQ v1.1

Our results suggest that the SAQ v1.1 is appropriate for clinical and research use in AIS. Additionally, the SAQ v1.1 appears more likely to detect changes in evaluative studies than the SAQ as its scores do not suffer from high ceiling effects (3% to 14%) and high floor effects (0 to 10%). A study by Schreiber et al. supports these findings by showing that the SAQ had a ceiling effect of >27% in adolescents who were treated using Schroth exercises.[42] A study by Thielsch T. et al.[77] featured findings that were comparable to our results, showing that the SAQ v1.1. had reliable test-retest measures (test-retest $r \geq 0.80$) and convergent validity with both the SRS-22r ($r = -0.40$ to -0.53) and Cobb angle ($r = 0.44$ to 0.55). It is worth noting that this study reported that the Expectations domain of the SAQ v1.1 did not demonstrate convergent validity with the Cobb angle ($r = 0.10$). However, this finding may be attributable to the fact that the questions related to

this domain concern expectations related to appearance of multiple body parts not only the chest or the breast .

Furthermore, in adults with scoliosis, the German version of the SAQ v1.1 correlated with both the Self-image domain and Total score of the SRS-22r (r = -0.53 and r = -0.60, respectively), supporting convergent validity. Along with our own findings, these results supported the decision to adopt this version as a reference score in our assessment of the new questionnaires.[77] Overall, we found that the SAQ v1.1 is better than the original SAQ for assessing convergent validity with new questionnaires in both adolescents and adults with scoliosis.

6.2.2 Measurement Properties of the New Questionnaires in Adolescents with Scoliosis

Chapters 4 and 5 present novel studies. To our knowledge, they are the first studies to evaluate the test-retest reliability and convergent validity of the new questionnaires against established questionnaires and radiographic measurements in adults and adolescents with scoliosis. Using metrological evaluation in the same population enables us to compare the assessments and characterize their performance in both adolescents and adults with scoliosis.

In Chapter 4, we concluded that the ISYQOL is more likely to detect changes in evaluative studies of the QOL than the SRS-22r in adolescents with IS because its score is free from ceiling and floor effects. The use of the ISYQOL of adolescents with idiopathic scoliosis was additionally supported by findings from a study conducted by Carroni et al., [142] where they concluded that the ISYQOL (the Italian version) has better known-groups validity and is more sensitive at detecting impact of disease severity on HRQOL in adolescents with scoliosis compared to the SRS-22.[142]

Our results suggest that the ISYQOL, TAASQ, and BIDQ-S questionnaires demonstrated an acceptable level of measurement reliability and convergent validity and can be used for research to assess patient perceptions regarding their appearance. Our findings also suggested that only the TAASQ Appearance and Clothing domains and subdomains met the reliability threshold for use in both clinical and research evaluations according to COSMIN. Lower reliability for some items and questionnaires may have resulted from having a focus on problems that are less relevant to adolescents with scoliosis. For example, questions related to domains on breast shape, size, and location may have been less relevant to younger adolescents and therefore generated results that were less reliable. Overall, this is one of the first studies to assess the validity and reliability of the TAASQ. However, a previous study on the BIDQ-S reported comparable findings to our BIDQ-S results, suggesting that the BIDQ-S has good internal consistency (Cronbach alpha = 0.82) and convergent validity compared to the SRS-22 Total score (r = -0.50 to -0.72) in patients with severe AIS.[141]

6.2.3 Measurement Properties of the New Questionnaires in Adults with Scoliosis

Similar to our study on adolescents with scoliosis, our results showed that the ISYQOL did not demonstrate convergence with radiograph measurements. In results found previously by our research team, we found that QOL became linearly related to Cobb angle measurements in a detectable manner only once severity exceeded 40°.[77,153] Since the ISYQOL was designed to capture the concerns of patients with mild and moderate scoliosis who are treated conservatively, its content may indeed have lower association with measurements of internal scoliosis severity that are not noticeable in terms of external appearance. Additionally, in adults, this may be

attributable to the content development of the ISYQOL as it was created using input from adolescent patients and their clinicians without consideration of the concerns expressed by adults with scoliosis.[144] For example, back pain is a less common complaint in patients with Adolescent Idiopathic Scoliosis compared to the adults with scoliosis where complaints regarding back pain affect 60 to 93% of the population and is usually associated with radicular leg pain and spinal stenosis.[28,4] Due to the fact that the ISYQOL only contains one question related to pain, asking about concerns of having pain in the future, it may be missing assessment of a component critical to QOL in adults with scoliosis.

The $ICC_{3.1}$ obtained for the ISYQOL was 0.89 in adolescents compared to the $ICC_{3.1}$ value of 0.70 found in adults. To our knowledge only one other study has examined measurement properties of the ISYQOL in adults. Zaina and colleagues found acceptable reliability ($\alpha = >0.7$) using internal consistency in adults with idiopathic scoliosis. Their study suggested that the translated ISYQOL could be a useful tool to measure quality of life in adults with scolisis.[82]

The Breast domain of the TAASQ demonstrated convergence only with the SAQ Total score and Appearance domain. This may be attributable to the pictograms provided in the SAQ, enabling patients to precisely describe their cosmetic issues. The TAASQ (except for its Breast location subdomain) and BIDQ-S (except for its Expectations domain) demonstrated convergent validity by significantly correlating with the SAQ v1.1. Similarly, the TAASQ (except for its Breast domain) and the BIDQ-S demonstrated convergent validity by significantly correlating with the SRS-22r. Additionally, convergence with Cobb angle was found only with the appearance-related domains of the TAASQ (Breast and Clothing domains). The results showing low convergence of

the TAASQ Appearance domain and Cobb angle may be due to the fact that most of the TAASQ questions are clothing and breast related. Furthermore, no significant correlations were found between lumbar rotation and any of the new questionnaires. This finding may be attributed to the soft tissue surrounding the lumbar spine, which may mask the effects of scoliosis on lumbar rotation and negatively perceived appearance. Additionally, amongst all of the adult participants, 22 presented with thoracic curves with an average Cobb angle of $42^{o} \pm 20^{o}$, while only 19 presented with lumbar curves with an average Cobb angle of $36^{o} \pm 15^{o}$. Thoracic vertebral rotation demonstrated convergence with only the TAASQ Breast domains and subdomains. This result supports the rationale that thoracic deformity is more likely to affect breast appearance than any other radiographic measurement.

On the basis of reliability, researchers interested in measuring perceived appearance in adults should use either the BIDQ-S or the TAASQ. Only the TAASQ Appearance domain, Breast domain, and Breast Size subdomains reached acceptable test-retest reliability levels supporting their use for individual evaluation according to COSMIN.[132] This larger reliability for the Breast domains in adults compared to adolescents with scoliosis supports our proposed explanation that questions or assessments that focus on problems that are less relevant for a specific age group result in lower measured reliability.

Of note, our results suggested that four domains of the TAASQ (the Appearance domain, Clothing domain, Clothing General subdomain, and Clothing Specific subdomain) can be used for both research and clinical assessment in adolescents with idiopathic scoliosis. However, in the adults, only three domains of the TAASQ (the Appearance domain, Breast domain, Breast Size

subdomain) can be used in both research and clinical assessment. We believe that the test-retest reliability for the adults could have been underestimated in our study due to the differences in data collection between our adult population and our adolescent population. To calculate reliability in our study, the adolescent participants completed questionnaires twice, once one-week following their initial clinic visit and once two-weeks following the initial clinic visit. Both times, questionnaires were completed after participants visited with their clinician. The data collection timeline differed in our adult population. Although our adult participants also completed questionnaires twice, they completed their first set prior to meeting with their clinician and one-week following their initial appointment. We believe that patient perceptions may be altered following discussions with their clinicians.[154] Consequently, these patients may answer questionnaires slightly differently between administrations, thereby decreasing test-retest reliability.

Overall, the Breast domains of the TAASQ had the largest floor effects in both populations. Given the mild severity of scoliosis in our sample, this finding may indicate that Breast items do not capture variability in issues related to severe breast appearance. This evidence suggests there is a need for more sensitive items to capture severe breast-related problems.

6.3 Outcome Measure Selection

Having multiple questionnaires for measuring the same construct can make the measurement selection process challenging. None of the questionnaires that we assessed in this thesis can be recommended for all situations. Therefore, we leave the selection decision to the individual

researcher or the clinician based on their unique needs. To inform the selection of outcome measures, one should consider the measurement limitations, the measurement context, and the decisions that will be made based on its result. An extensive explanation for each factor, with examples related to each new questionnaire, was discussed in the previous chapters (refer to Chapter 4 and 5). Overall, however, we suggest the use of the BIDQ-S as a short assessment to conduct for assessing both appearance and QOL; the ISYQOL as having enough sensitivity to track changes in QOL over time; the SAQ v1.1 as having better measurement properties than the SAQ; and the TAASQ as being able to capture anterior deformity in more detail than any other questionnaires and having acceptable reliability for clinical use (**Table 7-1 and 7-2**).

6.4 Limitations

Specific limitations of each study were discussed in the relevant chapters. Nevertheless, we will discuss the common limitations in this section and provide suggestions to address these limitations in the future directions section.

One common limitation of metrological studies is selection bias. We recruited from a single specialized clinic. Despite the fact that this clinic attracts patients from most of Northern Alberta along with neighbouring provinces, our recruitment from one clinic may affect the representativeness of our sample. However, many patients with mild symptoms typically do not seek medical assistance, thereby already limiting the generalizability of our results. The underrepresentation of mild and severe cases (we excluded patients who were planning to have

surgeries) may have lowered the variability of the sample which, as a result, may have lowered the reliability and affected correlations due to restrictions on range.[78]

The sample size for the adult study is another limitation. The final sample of 24 participants for our study on adult scoliosis was reduced from the initial recruitment goal of 100 participants. This reduction in number may have led to smaller sample variance in both QOL and perceived appearance responses. This restriction on the range may have resulted in an underestimation of test-retest reliability and decreased our ability to have precise confidence intervals. The difference in sample size between our adult population and our adolescent population may have affected our ability to detect differences in the performance of the questionnaires between the two populations. To increase our sample size, we contacted other hospitals in Calgary, Saskatchewan, and Vancouver to assist with the recruitment. However, we either did not receive a response from the site or the site declined recruitment in order to avoid creating additional burden on their patients given other ongoing studies. Finally, both the adult and adolescent studies planned for participants to complete questionnaires three times. However, the response rate of the second data collection time-point was interrupted by COVID-19, disrupting data collection in our adult population. In March of 2020, we decided to terminate the recruitment process due to concerns regarding the COVID-19 pandemic and the limited access that our participants faced in returning to our enrollment sites.

6.5 Future Directions

Future directions presented in this chapter are intended to build on the findings of the studies included in this dissertation. Our systematic review included only one study and showed limited evidence regarding specific target outcomes.[145] Therefore, to find additional literature, we recommend to researchers seeking to conduct systematic reviews on the effect of conservative treatment on adults with scoliosis to include studies conducted in other languages, studies using outcome measures such as Cobb angles, and studies on the effects of different therapeutic exercise on scoliosis. Additionally, to be more inclusive, future systematic reviews should include registries, theses, and abstracts. Soliciting studies known to experts in the field and identifying additional references from reference lists of included studies could also be performed in order to expand systematic reviews.

Measurement properties of the two SAQ versions were tested only in adolescent patients where we had a larger sample population. Measurement properties of the SAQ and SAQ v1.1 could also be analyzed in adults. To conduct studies on reliability, we suggest that future research evaluate other types of reliability, including internal consistency and reliability between paper administration vs computer-based administration vs the use of computer-adaptive questionnaires. Due to the fact that reliability estimates are sample dependant, to verify if higher reliability estimates are achievable with these questionnaires, future studies should have large and representative samples (more heterogeneous sample) to ensure high population variability during testing.[132] Ultimately, however, the population chosen to assess reliability should represent the group in which the questionnaire will be implemented. Future studies could also assess test-retest reliability over longer time-intervals as clinical follow up for scoliosis is typically conducted once

every 6 months. Furthermore, this thesis featured the use of Classical Test Theory (CTT); however, future researchers may consider the use of generalizability theory in order to identify the most important sources of score variation affecting repeatability.[132] Finally, researchers can use Bland-Altman plots to determine if error varies with the severity of the QOL or appearance issues. Additionally, because the COVID-19 pandemic limited access to medical services such as physiotherapy, research should be conducted on the validity and reliability of remote questionnaire administration (through phone calls, video conferencing, and other telehealth modalities).

For validity studies, we encourage researchers to include other radiograph measurements that correlate significantly with quality of life. For instance, patients with positive sagittal malalignment have reported worse self-assessment in pain, function, and self-image.[137] However, our studies did not include measurement of sagittal balance due to limitations in available technology and ethical concerns. Validation is an ongoing process, meaning that stronger evidence of validity can be accumulated over time based on the development of new theories that elaborate on relationships between construct under investigation and other constructs.[132] Therefore, future studies can evaluate other types of validity such as content, divergent, and known-group validity. It is not possible for studies evaluating the measurement properties of HRQOL to assess criterion validity due to the absence of a gold standard for this construct.

To have adequate measurements of QOL in adults with scoliosis, it might be helpful to review the content validity of the new questionnaires or to develop new questionnaires based on input from patients and other stakeholders such as therapists, surgeons, and other clinicians with expertise in adult scoliosis. New questionnaires should aim to cover all aspects of QOL specifically for adults

with scoliosis as most of the tools available to date have been developed for use primarily in adolescent populations.

Determining the ability of the questionnaires to detect change (i.e. responsiveness) is important, especially for evaluative studies. In the current research literature, SEM and MDC (i.e. distribution-based methods to measure responsiveness) are used to assess the ability of the SAQ v1.1, ISYQOL, TAASQ, and BIDQ-S to detect changes in adolescents and adults with mild to moderate scoliosis.[155] Future studies are recommended to evaluate the responsiveness of the questionnaires in more representative samples (i.e. mild, moderate, and severe scoliosis) depending on the planned application. Responsiveness studies using anchor-based methods are needed whereby changes in the tool under investigation is compared to a second, external measure of change that is more clearly understood to reflect judgement of the patient's or another stakeholder's perception of the importance of this change.[155]

6.6 Conclusions

Overall, this thesis highlighted the need for studies evaluating the effect of exercise on QOL in adults with scoliosis. Additionally, it suggested that clinicians and researchers should use the SAQ v1.1 instead of the old version of the SAQ. It also provides evidence of test-retest reliability and construct validity for the new questionnaires and introduces early sensitivity to change information required by researchers and clinicians to decide on which questionnaire is most adequate based on their situations in both adults and adolescents with scoliosis. In general, all questionnaires demonstrated convergent validity and adequate reliability for research use in both adults and

adolescent with scoliosis. However, four domains of the TAASQ (Appearance, Clothing domain, Clothing General subdomain, and Clothing Specific subdomain) can be used in both research and clinical care in adolescents with idiopathic scoliosis according to COSMIN standards. Whereas only three domains of the TAASQ (Appearance domain, Breast domain, and Breast Size subdomain) can be used in both research and clinical care in adults with scoliosis according to COSMIN standards.

Table 6-1: Summary of the Measurement Properties of the New Questionnaires in Adolescents with Idiopathic Scoliosis

	Test-retest reliability		Convergent Validity		Ceiling & Floor Effects	
	For Research ($ICC_{3.1} \geq 0.70$)	For Individuals ($ICC_{3.1} \geq 0.90$)	Established Questionnaires ($r \geq 0.35$)	Radiographic measurements ($r \geq 0.35$)	Ceiling (≤15%)	Floor (≤15%)
ISYQOL	✓	×	✓	×	✓	✓
TAASQ - Breast	✓	×	✓	✓	✓	×
TAASQ - Breast location	✓	×	✓	×	✓	×
TAASQ - Breast shape	✓	×	✓	✓	✓	×
TAASQ - Breast size	✓	×	✓	×	✓	×
TAASQ - Appearance	✓	✓	✓	✓	✓	✓
TAASQ - Clothing	✓	✓	✓	✓	✓	×
TAASQ- Clothing general	✓	✓	✓	✓	✓	×
TAASQ - Clothing Specific	✓	×	✓	×	✓	×
BIDQ-S Total	✓	×	✓	✓	×	✓

✓: Questionnaire/item met the minimum threshold:

For test-retest reliability: $ICC_{3.1} \geq 0.70$ for research use and $ICC_{3.1} \geq 0.90$ for individual use.

For convergent validity: correlation magnitude [$r \geq 0.35$] with at least one outcome measure [e.g. Cobb angle or SAQ v1.1 Total score])

For ceiling and floor effects: less than 15% of the participants scored the best or worst score.

×: Item did not meet the minimum threshold at all.

ISYQOL: Italian Spine Youth Quality of Life**, SRS-22r:** Scoliosis Research Society-22 (refined)**, SAQ V1.1:** Spinal Appearance Questionnaire version 1.1**, TAASQ:** Truncal Anterior Asymmetry Scoliosis Questionnaire**, BIDQ-S:** Body Image Disturbance Questionnaire-Scoliosis version, **ICC:** Intra-class Correlation Coefficient, and **r:** Pearson correlation coefficient

Table 6-2: Summary of the Measurement Properties of the New Questionnaires in Adults with Scoliosis

	Test-retest reliability		Convergent Validity		Ceiling & Floor Effects	
	For Research ($ICC_{3.1} \geq 0.70$)	For Individuals ($ICC_{3.1} \geq 0.90$)	Established Questionnaires ($r \geq 0.35$)	Radiographic measurements ($r \geq 0.35$)	Ceiling (≥15%)	Floor (≥15%)
ISYQOL	✓	×	✓	×	✓	✓
TAASQ - Breast	✓	✓	✓	✓	✓	×
TAASQ - Breast location	✓	×	✓	✓	✓	×
TAASQ - Breast shape	✓	×	✓	✓	✓	×
TAASQ - Breast size	✓	✓	✓	✓	✓	×
TAASQ - Appearance	✓	✓	✓	✓	✓	✓
TAASQ - Clothing	✓	×	✓	✓	✓	✓
TAASQ- Clothing general	✓	×	✓	✓	✓	✓
TAASQ - Clothing Specific	✓	×	✓	✓	✓	✓
BIDQ-S Total	✓	×	✓	✓	✓	✓

✓: Questionnaire/item met the minimum threshold:

For test-retest reliability: $ICC_{3.1} \geq 0.70$ for research use and ICC3.1 ≥ 0.90 for individual use.

For convergent validity: correlation magnitude [$r \geq 0.35$] with at least one outcome measure [e.g. Cobb angle or SAQ v1.1 Total score])

For ceiling and floor effects: less than 15% of the participants scored the best or worst score.

×: Item did not meet the minimum threshold at all.

ISYQOL: Italian Spine Youth Quality of Life, **SRS-22r:** Scoliosis Research Society-22 (refined), **SAQ V1.1:** Spinal Appearance Questionnaire version 1.1, **TAASQ:** Truncal Anterior Asymmetry Scoliosis Questionnaire, **BIDQ-S:** Body Image Disturbance Questionnaire-Scoliosis version, **ICC:** Intra-class Correlation Coefficient, and **r**: Pearson correlation coefficient

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Appendix 6-1: Ethics Approval

Notification of Approval (Renewal)

Date:	April 26, 2020
Amendment ID:	Pro00073569_REN3
Principal Investigator:	Eric Parent
Study ID:	MS6_Pro00073569
Study Title:	Translation to English, Reliabiilty and Validity Testing of the Italian Spine Youth Quality of Life (ISYQOL) questionnnaire.
Approval Expiry Date:	Friday, April 23, 2021

Thank you for submitting this renewal application. Your application has been reviewed and approved.

This re-approval is valid for another year. If your study continues past the expiration date as noted above, you will be required to complete another renewal request. Beginning at 30 days prior to the expiration date, you will receive notices that the study is about to expire. If you do not renew on or before the renewal expiry date, you will have to re-submit an ethics application.

All study related documents should be retained so as to be available to the Health REB upon request. They should be kept for the duration of the project and for at least 5 years following study completion.

Sincerely,

Charmaine Kabatoff, REB Consultant, for

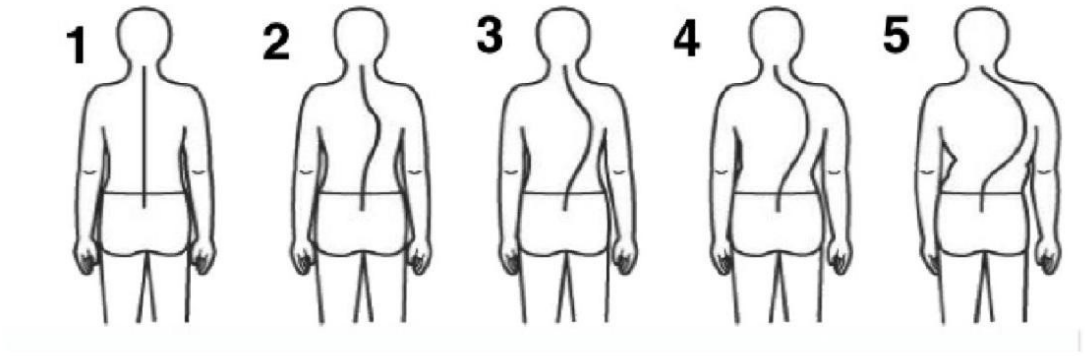
Anthony S. Joyce, PhD.
Chair, Health Research Ethics Board - Health Panel

Note: This correspondence includes an electronic signature (validation and approval via an online system).

Appendix 6-2 Established and New Questionnaire

Spinal Appearance Questionnaire (SAQ) Please look carefully at the following statements and pictures. Please select one that best describes your condition

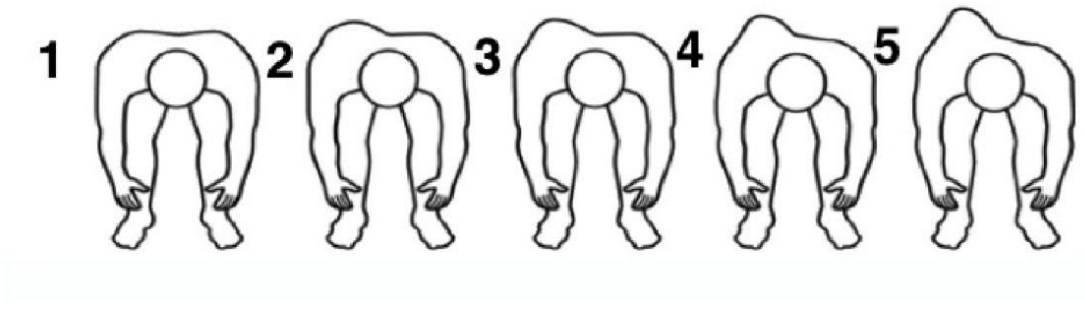
01. Body curve



Please select the number corresponding to the figure that best describes your condition

○ 1 ○ 2 ○ 3 ○ 4 ○ 5

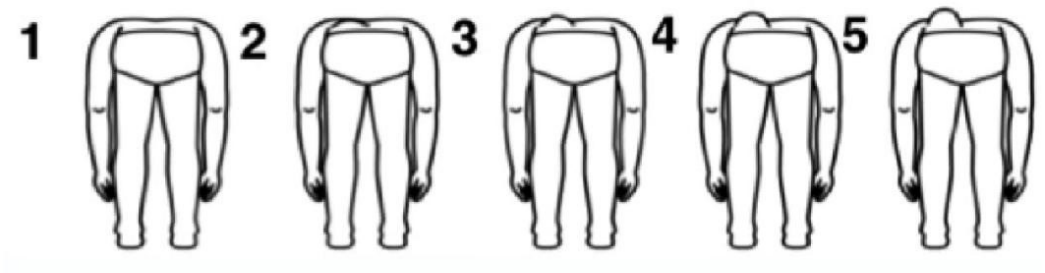
02. Rib prominence (bump)



Please select the number corresponding to the figure that best describes your condition

○ 1 ○ 2 ○ 3 ○ 4 ○ 5

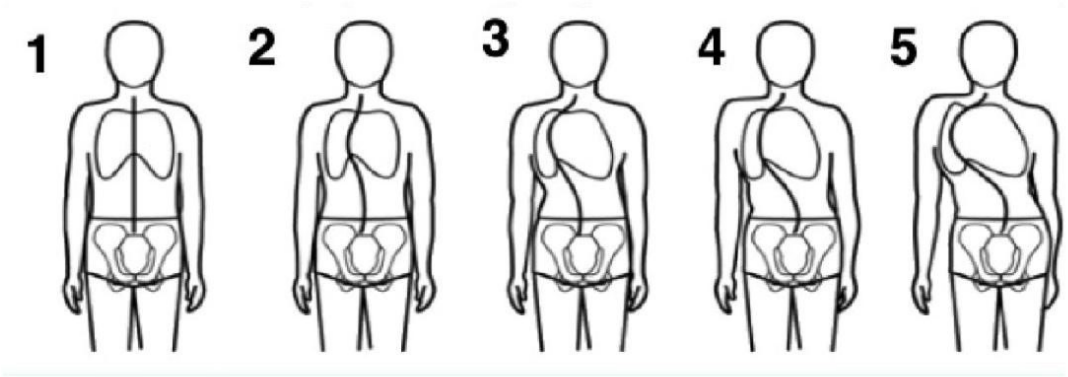
03. Flank prominence (bump)



Please select the number corresponding to the figure that best describes your condition

○ 1 ○ 2 ○ 3 ○ 4 ○ 5

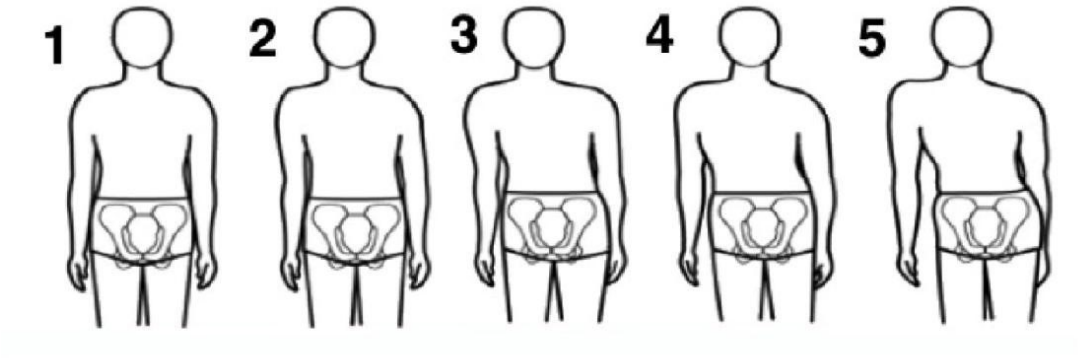
04. Head Chest Hips



Please select the number corresponding to the figure that best describes your condition

○ 1 ○ 2 ○ 3 ○ 4 ○ 5

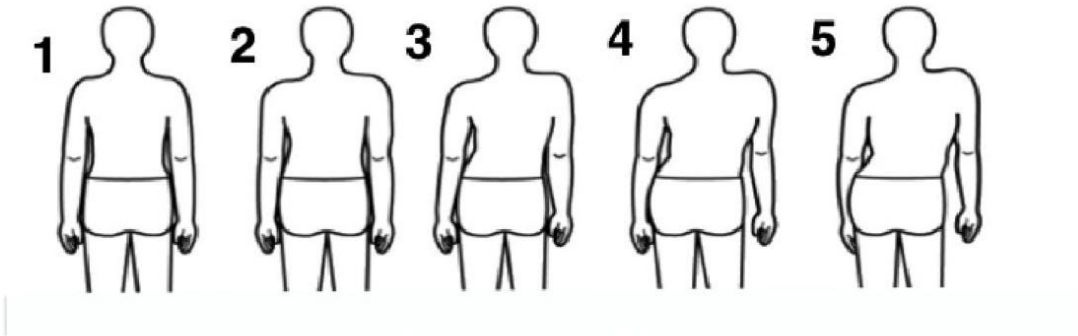
05. Position of head over hips



Please select the number corresponding to the figure that best describes your condition

○ 1 ○ 2 ○ 3 ○ 4 ○ 5

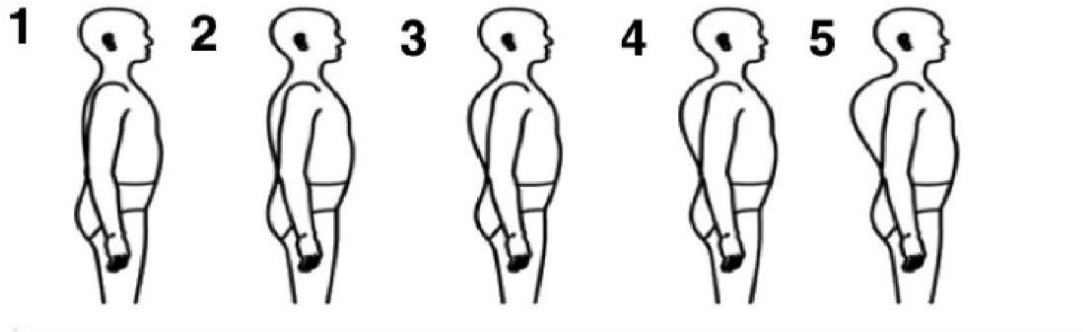
06. Shoulder level



Please select the number corresponding to the figure that best describes your condition

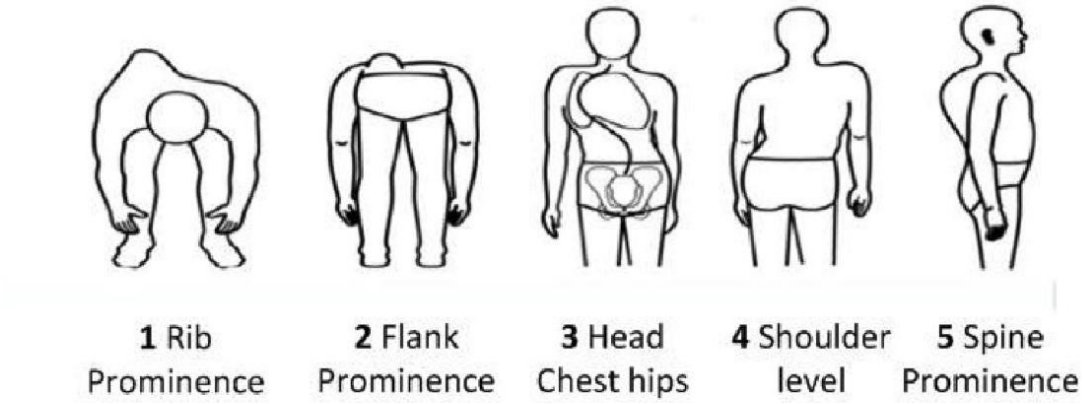
○ 1 ○ 2 ○ 3 ○ 4 ○ 5

07. Spine prominence (bump)



Please select the number corresponding to the figure that best describes your condition

◯ 1 ◯ 2 ◯ 3 ◯ 4 ◯ 5



08. Please, pick one category that bothers you the most out of these 5 categories of images

◯ 1 Rib prominence
◯ 2 Flank prominence
◯ 3 Head Chest Hips
◯ 4 Shoulder level
◯ 5 Spine prominence

How true is the following statement?

	Not true	A little true	Somewhat true	Fairly true	Very true
09. I want to be more even	◯	◯	◯	◯	◯
10. I want to look better in clothes	◯	◯	◯	◯	◯

11. I want to havemore even hips	○	○	○	○	○
12. I want to have a more even waist	○	○	○	○	○
13. I want to have more even leg length	○	○	○	○	○
14. I want to have more even breasts	○	○	○	○	○
15. I want to have a more even chest in the front	○	○	○	○	○
16. I want to have more even shoulders	○	○	○	○	○
17. I am self-conscious about my spine surgery scar	○	○	○	○	○

18. Of the questions 9 - 17, which one is the most important to you?

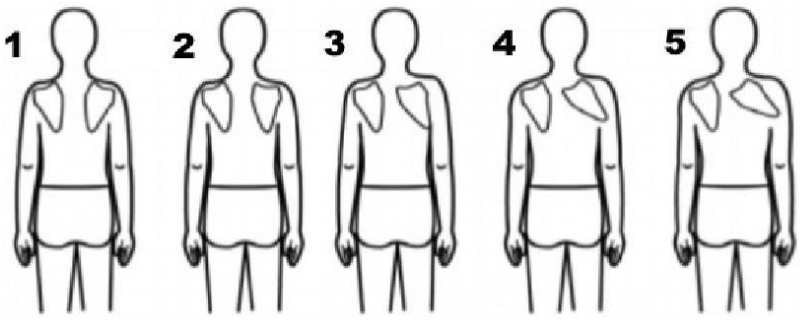
○ "I want to be more even"
○ "I want to look better in clothes"
○ "I want to have more even hips"
○ "I want to have a more even waist"
○ "I want to have more even leglength"
○ "I want to have more evenbreasts"
○ "I want to have a more even chest in the front"
○ "I want to have more even shoulders"
○ "I am self conscious about my spine surgery scar"

19. How would you rate your self-image?

○ Very bad ○ Bad ○ Fair ○ Good ○ Very good

Please look carefully at the following statements and pictures. Please select one that best describes your condition

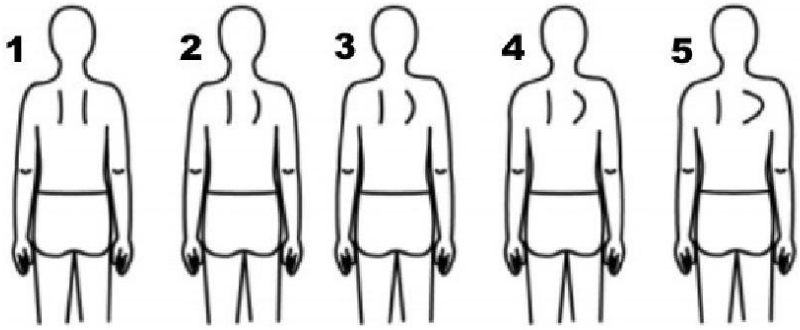
21. Shoulder blade rotation



Please select the number corresponding to the figure that best describes your condition

○ 1 ○ 2 ○ 3 ○ 4 ○ 5

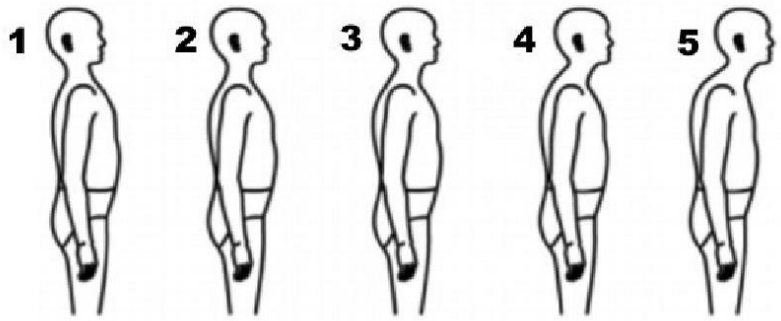
22. Shoulder angle



Please select the number corresponding to the figure that best describes your condition

○ 1 ○ 2 ○ 3 ○ 4 ○ 5

23. Head position



Please select the number corresponding to the figure that best describes your condition

○ 1 ○ 2 ○ 3 ○ 4 ○ 5

20. What would you most like to change about your body's shape and why?

Please type your answer in the field provided.

Scoliosis Research Society-22 (SRS-22) Please read the following questions carefully. Select one response that reflects your condition.

Today's Date	_____ ((mm/dd/yyyy))
1. Which one of the following best describes the amount of pain you have experienced during the past 6 months?	◯ None ◯ Mild ◯ Moderate ◯ Moderate to severe ◯ Severe
2. Which one of the following best describes the amount of pain you have experienced over the last month?	◯ None ◯ Mild ◯ Moderate ◯ Moderate to severe ◯ Severe
3. During the past 6 months have you been a very nervous person?	◯ None of the time ◯ A little of the time ◯ Some of the time ◯ Most of the time ◯ All of the time
4. If you had to spend the rest of your life with your back shape as it is right now, how would you feel about it?	◯ Very happy ◯ Somewhat happy ◯ Neither happy nor unhappy ◯ Somewhat unhappy ◯ Very unhappy
5. What is your current level of activity?	◯ Bedridden ◯ Primarily no activity ◯ Light labor and light sports ◯ Moderate labor and moderate sports ◯ Full activities without restrictions
6. How do you look in clothes?	◯ Very good ◯ Good ◯ Fair ◯ Bad ◯ Very bad
7. In the past 6 months have you felt so down in the dumps that nothing could cheer you up?	◯ Very often ◯ Often ◯ Sometimes ◯ Rarely ◯ Never
8. Do you experience back pain when at rest?	◯ Very often ◯ Often ◯ Sometimes ◯ Rarely ◯ Never
9. What is your current level of work/school activity?	◯ 100% normal ◯ 75%normal ◯ 50% normal ◯ 25% normal ◯ 0% normal

10. Which of the following best describes the appearance of your trunk; defined as the human body except for the head and extremities?	◯ Very good ◯ Good ◯ Fair ◯ Poor ◯ Very poor
11. Which one of the following best describes your pain medication use for back pain?	◯ None ◯ Non-narcotics weekly or less (e.g. aspirin, Tylenol, Ibuprofen) ◯ Non-narcotics daily ◯ Narcotics weekly or less (e.g. Tylenol III, Lorcet, Percocet) ◯ Narcotics daily
12. Does your back limit your ability to do things around the house?	◯ Never ◯ Rarely ◯ Sometimes ◯ Often ◯ Very often
13. Have you felt calm and peaceful during the past 6 months?	◯ All of the time ◯ Most of the time ◯ Some of the time ◯ A little of the time ◯ None of the time
14. Do you feel that your back condition affects your personal relationships?	◯ None ◯ Slightly ◯ Mildly ◯ Moderately ◯ Severely
15. Are you and/or your family experiencing financial difficulties because of your back?	◯ Severely ◯ Moderately ◯ Mildly ◯ Slightly ◯ None
16. In the past 6 months have you felt downhearted and blue?	◯ Never ◯ Rarely ◯ Sometimes ◯ Often ◯ Very often
17. In the last 3 months have you taken any days off work, including household work or school, because of back pain?	◯ 0 days ◯ 1 day ◯ 2 days ◯ 3 days ◯ 4 or more days
18. Does your back conditions limit your going out with friends/family?	◯ Never ◯ Rarely ◯ Sometimes ◯ Often ◯ Very often
19. Do you feel attractive with your current back condition?	◯ Yes, very ◯ Yes, somewhat ◯ Neither attractive nor unattractive ◯ No, not very much ◯ No, not at all

20. Have you been happy person during the past 6 months?	◯ None of the time ◯ A little of the time ◯ Some of the time ◯ Most of the time ◯ All of the time
21. Are you satisfied with the result of your back management?	◯ Very satisfied ◯ Satisfied ◯ Neither satisfied nor unsatisfied ◯ Unsatisfied ◯ Very unsatisfied
22. Would you have the same management again if you had the same condition?	◯ Definitely yes ◯ Probably yes ◯ Not sure ◯ Probably not ◯ Definitely not

Spine Youth Quality of Life (SYQOL) Measuring Spine Related Quality of LifeWe want to evaluate your well-being with regards to your back problem (scoliosis, kyphosis or something else). Try to answer all of the following questions yourself.

1. Are you afraid that your back problem may get worse?	◯ never ◯ sometimes ◯ often
2. Are you worried about having back pain in the future because of your back problem?	◯ never ◯ sometimes ◯ often
3. Do you feel that having your back problem is a big deal?	◯ never ◯ sometimes ◯ often
4. Are you worried that, despite all your efforts to treat your back, it will not get better?	◯ never ◯ sometimes ◯ often
5. Do you think that your back problem is more serious than other health conditions affecting other people?	◯ never ◯ sometimes ◯ often
6. Despite your back problem, do you think you lead a normal life?	◯ never ◯ sometimes ◯ often
7. Are you suffering because of your back problem?	◯ never ◯ sometimes ◯ often
8. Does the appearance of your back make you feel uncomfortable?	◯ never ◯ sometimes ◯ often
9. Are you worried about your back problem?	◯ never ◯ sometimes ◯ often
10. Do you think that your back problem is not a big concern to you?	◯ never ◯ sometimes ◯ often
11. Does it bother you to show your physical appearance?	◯ never ◯ sometimes ◯ often
12. Are you worried that your back problem is very visible?	◯ never ◯ sometimes ◯ often
13. Despite your back problem, do you live a happy life?	◯ never ◯ sometimes ◯ often
Do you wear a brace because of your back problem?	◯ Yes ◯ No
14. Do you have to change the way that you dress because of your brace?	◯ never ◯ sometimes ◯ often

15. Are you worried that the brace is visible under your clothing?	◯ never ◯ sometimes ◯ often
16. Do you feel sad that you are unable to do some of the things that you used to do before you started wearing your brace?	◯ never ◯ sometimes ◯ often
17. Do you feel your movements are restricted while wearing your brace?	◯ never ◯ sometimes ◯ often
18. Does wearing your brace ever make you cry?	◯ never ◯ sometimes ◯ often
19. Do you feel excluded by others because you wear your brace?	◯ never ◯ sometimes ◯ often
20. Is wearing your brace uncomfortable?	◯ never ◯ sometimes ◯ often

If you would like, use this space to leave a comment.

Adolescent Idiopathic Scoliosis- Truncal Anterior Asymmetry Scoliosis Questionnaire
Directions: This questionnaire will ask you how you feel about your front body appearance. Please answer each of the following questions by marking the choice that best represents what you think. It is important to answer these questions by yourself.

1. How do you think you look in clothes?
- ◯ Very bad
- ◯ Bad
- ◯ Fair
- ◯ Good
- ◯ Very good

2. How do you think your following body parts look in clothes?

	Very bad	Bad	Fair	Good	Very good
Front trunk	◯	◯	◯	◯	◯
Breasts	◯	◯	◯	◯	◯
Front rib hump	◯	◯	◯	◯	◯
Hips	◯	◯	◯	◯	◯
Waist	◯	◯	◯	◯	◯

3. How do you think you look from the front, unclothed?
- ◯ Very bad
- ◯ Bad
- ◯ Fair
- ◯ Good
- ◯ Very good

4a. On a scale of 1 to 5, 1 meaning and 5 meaning, how would you rate your breasts? Would you say that your breasts are matched:

	1 very different	2	3	4	5 very matched
In size?	◯	◯	◯	◯	◯
In shape?	◯	◯	◯	◯	◯
In location?	◯	◯	◯	◯	◯

4b. If you answered anything other than to any of the above, please describe any issue that bothers you about the appearance of your breasts. Please be as specific as possible.

_____

5. How often do you think about your front appearance?
- ◯ Never
- ◯ Rarely
- ◯ Sometimes
- ◯ Often
- ◯ Very often

6. If you think any or many parts of your front body (such as breasts, ribs, hips, waist) are unmatched, how much does this bother you?
- ◯ Not at all
- ◯ Slightly
- ◯ Mildly
- ◯ Moderately
- ◯ Severely

7a. How much would you say you have changed your daily life activities (eating, dressing, shopping, walking etc.) as a result of your front body appearance?
- ◯ Not at all
- ◯ Slightly
- ◯ Mildly
- ◯ Moderately
- ◯ Severely

7b. What changes have you made in your activities of daily life for this reason? Please specify which body part(s) these modifications in your life are related to. Elaborate as much as possible.

_____

8a. When you choose the clothes you are going to wear, do you avoid:

	Never	Rarely	Sometimes	Often	Very often
Certain types of clothing?	○	○	○	○	○
Certain types of undergarments (bras or underwear)?	○	○	○	○	○
Certain types of bathing suits?	○	○	○	○	○
Other	○	○	○	○	○

If you answered the category, please specify

_____

8b. If you answered anything other than to any of the above,, please list the types of clothing and explain why you avoid them. Please be as specific as possible.

_____

9a. Do you use clothing to hide your body shape?

- ○ Never
- ○ Rarely
- ○ Sometimes
- ○ Often
- ○ Very Often

9b. If so, which body parts and why? Please elaborate as much as possible.

_____

10. When you think about your front appearance, do you feel different from other girls?

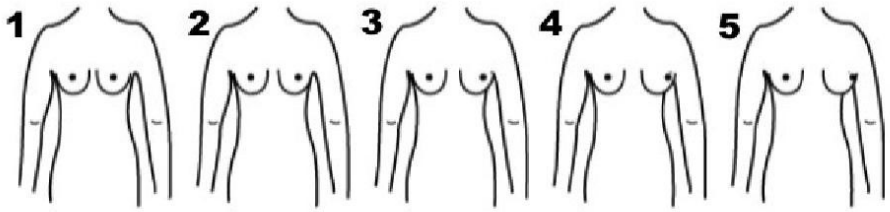
- ○ Not at all
- ○ Slightly
- ○ Mildly
- ○ Moderately
- ○ Severely

11. If you had to spend the rest of your life with your current chest, ribs, waist, and hips as they are right now, how would you feel about it?

- ○ Very bad
- ○ Bad
- ○ Fair
- ○ Good
- ○ Very good

Directions: Please look carefully at the following pictures. Please choose the figure that best describes your condition. Do not take into account if the unmatched side in the picture is different from yours. Just answer based on the amount of difference between the right and left sides of your body.

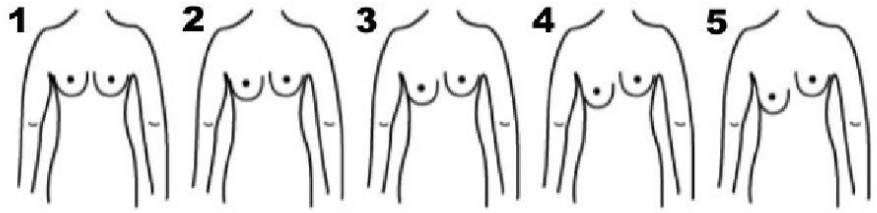
12. Horizontal location of breasts



Please select the number corresponding to the figure that best describes your condition

○ 1 ○ 2 ○ 3 ○ 4 ○ 5

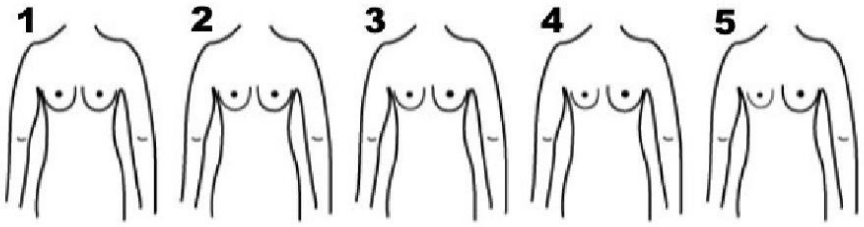
13. Vertical location of breasts



Please select the number corresponding to the figure that best describes your condition

○ 1 ○ 2 ○ 3 ○ 4 ○ 5

14. Size of breasts compared to each other



Please select the number corresponding to the figure that best describes your condition

○ 1 ○ 2 ○ 3 ○ 4 ○ 5

Please comment on any questions of the questionnaires which are unclear or need revision (if possible, refer to the variable name)

You can leave the questionnaire and return to complete it later. To save your answer and be able to continue where you have left off, you have to scroll to the bottom of the page and click the button "Save and Return later".

If you leave the survey, a code with be generated automatically for you. Please take note of it. To return and complete where you left off you will need your code to access the survey again. You have a maximum of 7 days to complete the questionnaire if you decide to return.
Thank you!

BODY IMAGE DISTURBANCE QUESTIONNAIRE-SCOLIOSIS

Kids and teenagers with scoliosis sometimes have concerns about their appearance, or how they look. This questionnaire asks about concerns you may have about how your back looks because you have scoliosis.

Please read each question carefully and circle the answer that best describes your thoughts and feelings about your back. Some questions ask you to write in your own answers. Please write your answers on the line that is provided. There are no right or wrong answers.

1A. Are you worried about the appearance of your back shape?

- ◯ Not at all worried
- ◯ Somewhat worried
- ◯ Moderately worried
- ◯ Very worried
- ◯ Extremely worried

1B. What are these concerns? (Check all that apply)

- ☐ My shoulders are uneven (one is higher or lower than the other)
- ☐ My shoulder blade sticks out
- ☐ My chest is asymmetric from the front (one side looks higher or lower than the other side)
- ☐ My hips are asymmetric (one hip is higher or lower than the other)
- ☐ My rib bump

2A. If you are at least somewhat concerned or worried, do these concerns/worries preoccupy you? That is, do you think about them a lot and are they hard to stop thinking about?

- ◯ Not at all preoccupied (I do not think about them)
- ◯ Somewhat preoccupied (I think about them from time to time)
- ◯ Moderately preoccupied (I think about them a moderate amount)
- ◯ Very preoccupied (I think about them a lot)
- ◯ Extremely preoccupied (I think about them constantly)

2B. How do your concerns about the way your back looks affect your life? For example, some kids say that they avoid swimming because they are embarrassed to show their back.

_____

3. Has the way your back looks caused you to feel upset? How much?

- ◯ Not upset at all
- ◯ Mild (a little bit upset)
- ◯ Moderate (Somewhat upset)
- ◯ Severe, and very disturbing (very upset)
- ◯ Extreme, and disabling (extremely upset)

4. Have your worries about how your back looks caused you any problems at school, at your job, or with your friends and family? How much?

- ◯ No problems
- ◯ A few problems, but overall I can do what I need to do, and my performance is not affected
- ◯ Several problems, but I can cope with them; problems are still manageable
- ◯ A lot of problems that limit what I can do; problems cause a lot of limitations
- ◯ Extreme problems that keep me from doing almost everything I want or need to do

5A. Has your back shape caused problems with your friends, family members, or dating? How much?	◯ Never ◯ Occasionally ◯ Sometimes ◯ A lot ◯ All the time
5B. If so, how?	_____
6A. Has your back shape caused problems with your schoolwork, your job, or your ability to do other things that are important to you (e.g., play sports, be social with your friends)? How much?	◯ Never ◯ Occasionally ◯ Sometimes ◯ A lot ◯ All the time
6B. If so, how?	_____
7A. Do you ever avoid things because of your back shape? How often?	◯ Never ◯ Occasionally ◯ Sometimes ◯ A lot ◯ All the time
7B. If so, what things do you avoid?	_____

Appendix 6-3: Systematic Review – Search Strategies

Ovid MEDLINE

(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily and Ovid MEDLINE(R) 1946 to Present

Results: 209

Date: March 9, 2017

1. spinal curvatures/ or scoliosis/

2. (Scoliosis or scoliotic or (((de novo or degenerative) adj4 (deformit* or curve* or curvature*)) and (spine or spinal or vertebral)) or ((spine or spinal) adj4 deformit*)).mp.

3. 1 or 2

4. exercise therapy/ or plyometric exercise/ or resistance training/

5. Exercise/

6. (muscle strengthen* or (motor adj4 rehab) or ((task orient* or stabili* or core or abdominal or multifidus or strength* or motor control or weight or resist*) adj4 (train* or exercise*)) or pilates or abdominal bracing or "contralateral arm lift*" or sit up* or sit-up* or situp* or crunches or "draw* in maneuver*" or "draw* in manoeuvre*").mp.

7. 4 or 5 or 6

8. 3 and 7

9. limit 8 to English language

10. limit 9 to (comment or editorial or letter)

11. 9 not 10

12. 11 not ((child or children) not adult*).ti.

13. remove duplicates from 12

EBM Reviews - Cochrane Central Register of Controlled Trials January 2017

Date searched: March 9, 2017

Results: 29

1. spinal curvatures/ or scoliosis/

2. (Scoliosis or scoliotic or (((de novo or degenerative) adj4 (deformit* or curve* or curvature*)) and (spine or spinal or vertebral)) or ((spine or spinal) adj4 deformit*)).mp.

3. 1 or 2

4. exercise therapy/ or plyometric exercise/ or resistance training/

5. Exercise/

6. (muscle strengthen* or (motor adj4 rehab) or ((task orient* or stabili* or core or abdominal or multifidus or strength* or motor control or weight or resist*) adj4 (train* or exercise*)) or pilates or abdominal bracing or "contralateral arm lift*" or sit up* or sit-up* or situp* or crunches or "draw* in maneuver*" or "draw* in manoeuvre*").mp.

7. 4 or 5 or 6

8. 3 and 7

9. limit 8 to english language

10. limit 9 to (comment or editorial or letter)

11. 9 not 10

12. 11 not ((child or children) not adult*).ti.

13. remove duplicates from 12

Embase

Date:1974 to March 9, 2017

Results: 467

1. exp scoliosis/

2. (Scoliosis or scoliotic or (((de novo or degenerative) adj4 (deformit* or curve* or curvature*)) and (spine or spinal or vertebral)) or ((spine or spinal) adj4 deformit*)).mp.

3. 1 or 2

4. kinesiotherapy/ or muscle training/ or plyometrics/

5. resistance training/ or exercise/

6. (muscle strengthen* or (motor adj4 rehab) or ((task orient* or stabili* or core or abdominal or multifidus or strength* or motor control or weight or resist*) adj4 (train* or exercise*)) or pilates or abdominal bracing or "contralateral arm lift*" or sit up* or sit-up* or situp* or crunches or "draw* in maneuver*" or "draw* in manoeuvre*").mp.

7. 4 or 5 or 6

8. 3 and 7

9. limit 8 to english language

10. limit 9 to (editorial or letter)

11. 9 not 10

12. 11 not ((child or children) not adult*).ti.

13. remove duplicates from 12

14. limit 13 to conference abstract

15. 13 not

CINAHL

Date searched: March 9, 2017

Results: 167

	S11	s10 not TI ((child or children) not adult*) Limiters - English Language
	S10	S8 not s9
	S9	PT (comment or editorial or letter)
	S8	s3 and s7
	S7	s4 or s5 or s6
	S6	(muscle strengthen* or (motor n4 rehab) or ((task orient* or stabili* or core or abdominal or multifidus or strength* or motor control or weight or resist*) n4 (train* or exercise*)) or pilates or abdominal bracing or "contralateral arm lift*" or sit up* or sit-up* or situp* or crunches or "draw* in maneuver*" or "draw* in manoeuvre*")
	S5	(MH "Exercise")
	S4	(MH "Therapeutic Exercise") OR (MH "Abdominal Exercises") OR (MH "Back Exercises") OR (MH "Plyometrics") OR (MH "Muscle Strengthening") OR (MH "Resistance Training")
	S3	s1 or s2
	S2	(Scoliosis or scoliotic or (((de novo or degenerative) n4 (deformit* or curve* or curvature*)) and (spine or spinal or vertebral)) or ((spine or spinal) n4 deformit*))
	S1	(MH "Spinal Curvatures") OR (MH "Scoliosis+")

SportDiscus

Date searched: March 9, 2017

Results: 36

((Scoliosis or scoliotic or (((de novo or degenerative) n4 (deformit* or curve* or curvature*)) and (spine or spinal or vertebral)) or ((spine or spinal) n4 deformit*))) AND ((muscle strengthen* or (motor n4 rehab) or ((task orient* or stabili* or core or abdominal or multifidus or strength* or motor control or weight or resist*) n4 (train* or exercise*)) or pilates or abdominal bracing or "contralateral arm lift*" or sit up* or sit-up* or situp* or crunches or "draw* in maneuver*" or "draw* in manoeuvre*")) NOT (TI ((child or children) not adult*))