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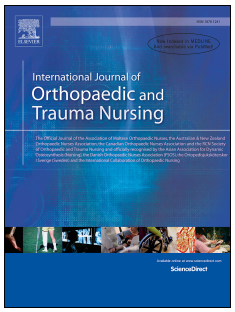


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# A systematic scoping review and textual narrative synthesis of long-term health-related quality of life outcomes for adolescent idiopathic scoliosis

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# A systematic scoping review and textual narrative synthesis of long-term health-related quality of life outcomes for adolescent idiopathic scoliosis


## Abstract

### Introduction

Idiopathic scoliosis is a musculoskeletal condition leading to deformity of the spinal column. There is a strong evidence reporting short term health-related quality of life outcomes, but less is known about the longer-term impact of adolescent idiopathic scoliosis (AIS). This paper reports the current evidence on long-term non-clinical outcomes of AIS.

### Method

A systematic scoping literature review, combining descriptive and textual narrative synthesis was undertaken. Studies were included if they sampled or followed up participants at least 10 years after diagnosis and / or treatment, contained health-related quality of life data that could be extracted, where the intervention (or diagnosis in the case of untreated) occurred after 1980 and where data was extractable for modern rod and screw or fusion techniques, non-surgical interventions or untreated patients.

### Results

Twenty-three studies were included. Overall, the HRQOL measures utilised by these studies suggest that HRQOL is not related to participant demographic or AIS characteristics or type or extent of surgical intervention. Some studies suggest that those with AIS scored worse than controls.

### Discussion

Results suggest that AIS participants had a generally good quality of life, although this was often worse than those without AIS. No other clear relationships were found. Importantly, the available literature fails to address more fundamental questions about how HRQOL is conceptualised for those with AIS, and there is value in pursuing qualitative inquiry in this area.

## Introduction

Idiopathic scoliosis is a musculoskeletal condition resulting in deformity of the spine, when unequal development of spinal musculature causes thoracic, lumbar or thoracolumbar curvatures and twists. It has a prevalence rate of 0.47 – 5.2% (Erwin et al., 2020; Konieczny et al., 2013). Although idiopathic scoliosis occurs in all age groups, 90% of cases are diagnosed in adolescents. The cause of Adolescent Idiopathic Scoliosis (AIS) is unknown, although genetic, embryological and developmental theories are proposed. Since the peak age of onset is puberty and the condition is more common in girls than in boys, there is much interest in the role of hormones in instigating scoliosis. Whilst usually asymptomatic, advanced curvatures can cause functional difficulties relating to:

a. respiration and digestion, as thoracic and abdominal compartments are compromised (Durmala et al., 2008)
b. pain and discomfort due to over-stretching of muscles and connective tissues on the external edge of the curves, and compression of the same structures on the internal edge (Makino et al., 2015)
c. impingement of nerves and nerve roots exiting the spine
d. height and gait, and
e. disfigurement, disrupting body image and self-esteem (Leszczewska et al., 2012)

Scoliosis sometimes responds to external bracing, which aims to support the spinal column back into the correct position, holding it in place to permit stretching of the tight muscles on the inner face of the curve(s). For unresponsive curvatures, spinal fusion surgery with internal permanent fixation is the optimal treatment. The decision to offer surgery is based mainly on the extent of the curvatures (Cobb angles), the age of the individual, and the rate of progression of curvature. Cobb angles of 50 degrees or more in the thoracic spine or 40 degrees or more in the lumbar spine are usually indicative of the need for surgery (Bettany-Saltikov et al., 2015).

Extensive research has guided refinement of best surgical practices. However, it is major invasive surgery, requires teams of highly skilled clinicians, and carries a 1 in 500 (less than 1%) risk of paralysis. Patients are often understandably nervous about agreeing to such

surgery, Despite risks and costs, clinical outcomes are usually excellent with very low complication rates, although as scoping/anecdotal evidence from online support platforms demonstrate, there are numerous long-term psychosocial concerns and daily challenges facing patients long after their surgery is carried out.

## Background

There is a substantial body of evidence that examines surgical and non-surgical outcomes in AIS, yet the literature on quality of life is more mixed. Pellegrino & Avanzi (2014) for example concluded that amongst 33 patients at 3, 6 and 12 months follow up, surgical treatment of AIS improved patient quality of life. In a systematic review, Rushton & Grevitt (2013) concluded that while no clinically relevant changes occurred in mental health or activity domains, surgery could lead to clinically important improvement in patient self-image. These results however contrast with other studies that suggest significant numbers of AIS patient face "clinically significant psychological symptoms" (Sanders et al., 2018).

Over the longer term, while there is a growing literature on outcomes of mid to long-term surgical and non-surgical outcomes of scoliosis (Kepler et al., 2012; Lykissas et al., 2013), there is far less evidence on the long-term social, psychosocial, health and economic outcomes amongst those with idiopathic scoliosis who do or do not have fusion surgery, making it difficult for these often young patients and their parents to make informed decisions about treatment and management of their condition.

Anecdotal evidence gathered by the research team, either through publicly available social media platforms or personal contact with people with fused or unfused idiopathic scoliosis clearly indicates the profound impact the condition has on individuals' lives. There are personal reports from those living with fused and unfused AIS, of living permanently with chronic pain, a reliance on often multiple analgesics, and disrupted ability to work due to fatigue, pain, or discomfort. Physical impacts include difficulties with maintaining any single position (sitting, walking, standing) for periods of time, and finding it hard to breath. There is an often unannounced yet apparent, through the nature of online posts and discussions, impact on mental health – with indications of anxiety, distress, and depression. For those with unfused scoliosis, another common emotion in online forums is a sense of hopelessness, since without the offer of surgery, there is no permanent solution to the curves.

The aim of this review was to summarise and synthesise the recent literature on the long-term (10 years+) health-related quality of life (HRQOL) outcomes for people with adolescent idiopathic scoliosis, who did or did not have modern (late 1980's onwards) spinal fusion surgery. The objective was to demonstrate that there is minimal evidence of psychosocial influences on HRQoL in this population. The review focused on the type of outcomes examined, and how these were measured. The research question was: What are the long-term non-clinical outcomes for patients with AIS who have or have not had spinal fusion surgery?' In presenting the narrative synthesis, the populations that have been researched, methodologies used, the shortcomings of this literature and recommendations for future policy and research are also discussed.

## Method

### Design

A systematic scoping review was utilised to examine the extent, range and nature of research activity and to identify gaps in the existing literature, with the goal of identifying, appraising, and synthesizing all relevant studies on the long term HRQOL outcomes of AIS (Uman, 2011), rather than generate a single outcome of interest. Pham et al. (2014) confirms that a scoping review, conducted in an organised (systematic) manner 'maps the existing literature … in terms of the volume, nature and characteristics of the primary research.' The scoping review reported here involved seven steps: 1) formulating a review question; 2) developing a search strategy and conducting the search; 3) defining inclusion and exclusion criteria; 4) selecting studies; 5) appraising included studies; 6) extracting data; and 7) analysing, synthesising and interpreting results. This review follows a results-based convergent synthesis design: qualitative, quantitative and mixed-methods studies were identified in a single search, integrated throughout analysis, synthesis and presentation (Hong et al., 2017; Noyes et al., 2019). PRISMA and ENTREQ reporting guidelines have been followed (Moher et al., 2009; Tong et al., 2012).

### Search Methods

Five reference databases were searched: Medline, CINAHL, Web of Science, Science Direct, and Scopus. Preliminary search terms were developed to reflect the core concepts - the

population of interest (those diagnosed with AIS), the outcomes or impact of treatment and the time frame of interest (long-term). The final search terms used were ("adolescent idiopathic scoliosis" OR "idiopathic scoliosis") AND (outcomes or benefits or effects or impact or effectiveness) AND ("long term" or long-term or longitudinal). The final search was conducted in June 2020. Results were collated and duplicate articles were removed.

### Inclusion/exclusion criteria

A total of 77 studies were identified, one full text was unobtainable (author emailed) and the remainder were reduced to 18 after being assessed against the following inclusion/exclusion criteria:

Inclusion

- Studies that sampled or followed up participants at least 10 years (mean) after intervention (or diagnosis in the case of those who did not undergo treatment)
- Studies that contained health-related quality of life data that could be extracted
- Studies where the intervention (or diagnosis in the case of untreated) for the entire sample occurred after 1980
- Studies where data was extractable for modern rod and screw or fusion techniques, non-surgical interventions or untreated AIS
- Studies of any design – quantitative, qualitative, mixed methods

Exclusion

- Studies that did not include extractable HRQOL data (i.e. studies that only examined radiographic results or surgical outcomes for example)
- Studies that examined traditional surgical techniques, including Harrington rod surgery
- Studies that were not available in English
- Conference abstracts
- Articles that were not published in peer reviewed journals

Following screening, studies were explored for further references that were not identified in the original search and that met the above criteria. A further five articles were identified, leaving 23 articles to be included in the review and analysis (Figure 1).

### Data extraction

Data was extracted by one author (RE) and categorised according to the source, country of where the research took place, study aims and objectives, research methods/design and sample information, participants, scales of analysis, main outcomes, and quality appraisal scores and issues (see Table 1).

### Quality appraisal

Critical Appraisal Skills Program (CASP) tools were adopted to assess the quality of selected publications. The CASP tools provide unique checklists for evaluating eight different types of studies. In this systematic review, the CASP cohort and case control study checklists were adopted (Critical Appraisal Skills Programme, 2018a, b). These contain 12, and 10 dichotomous response questions respectively. Both also include two questions summarising study results which were not utilised. Studies were therefore scored against a possible total score of 12 for cohort and 10 nine for cross sectional studies. To assess the quality of eligible studies, two authors (RE and LD) scored each study against its respective CASP criteria.

### Data summary and synthesis

Due to heterogeneity across studies and even within similar study methodologies, a meta-analysis or combining of quantitative data for further analysis, or a meta-synthesis for qualitative data was not possible. Instead studies were combined to summarise descriptive statistics of the study characteristics, followed by a textual narrative synthesis. This approach arranges disparate study types into more homogenous sub-groups which aids in the synthesising of different types of evidence. Study characteristics, context, quality, and findings are reported according to a standard format, and similarities and differences are compared across studies (Lucas et al., 2007).

## Results

### Quality appraisal results

Of the 23 included studies, two were scored against the CASP case control checklist (Enercan et al., 2015; Remes et al., 2004), while the remainder were scored against the CASP cohort study checklist. One, a case study, was excluded from quality appraisal (Lebel & Lebel, 2016). The two case control studies were rated as high quality, with each meeting eight out of

the ten possible quality indicators. Similarly, all cohort studies were of generally good quality. Only one study scored seven, with all others scoring eight or above. However, almost every cohort study failed to account for a range of potentially confounding variables in their design and analysis.

### Combined study descriptive results

All 23 papers utilised a quantitative approach, and overwhelmingly a cohort or case-control design. The research took place across twelve geographic locations. Six studies were carried out in the US (Erwin et al., 2020; Green et al., 2011; Kelly et al., 2010; Lavelle et al., 2016; Louer et al., 2019; Newton et al., 2020), four in Japan (Kino et al., 2019; Sudo et al., 2013a; Sudo et al., 2013b; Sudo et al., 2016), two in Switzerland (Boos et al., 2007; Min et al., 2013) two in Germany (Mueller & Gluch, 2009, 2012), two in Turkey (Benli et al., 2007; Enercan et al., 2015), and one in Norway (Bjerkreim et al., 2007), Canada (Lebel & Lebel, 2016), Finland (Remes et al., 2004), Israel (Falick-Michaeli et al., 2015), Italy (Scaramuzzo et al., 2017), Spain (Pérez-Grueso et al., 2000) and Sweden (Danielsson et al., 2010). The combined sample size was 1,559 (including a small number of controls). Twenty studies examined surgical outcomes, with the remaining three studies examining outcomes of brace and no treatment (Danielsson et al., 2010), no treatment (Erwin et al., 2020) or exercise (Lebel & Lebel, 2016). While the earliest study was conducted in the year 2000, the majority were undertaken in the last decade, since 2010.

### Textual narrative synthesis results

The included studies were categorized in relation to the health-related quality of life outcomes they assessed. The majority (n=21) of studies utilized various versions of the Scoliosis Research Society (SRS) questionnaire (Haher et al., 1999). Seven studies utilized the Oswestry Disability Index (ODI) (Fairbank & Pynsent, 2000). Three studies used the Short Form Survey (SF-36) (Ware et al., 1980). One used the Roland-Morris Disability questionnaire (Roland & Morris, 1983). One study used the EuroQol (EQ5D) (Balestroni & Bertolotti, 2012), one used the Beck Depression Inventory (Beck et al., 1996), a further study utilised the Female Sexual Distress Scale (FSDS) (Derogatis et al., 2002) and finally, one study used bespoke researcher-developed scales (Boos et al., 2007). The studies that utilised each of these measures, along with their key findings are summarised in Table 2.

*Scoliosis Research Society results*

The SRS has a number of variants (SRS-22, 23, 24 and 30) and has been translated into several languages. While items differ slightly across versions, the SRS contains questions that examine five domains: function/activity, pain, self-image/appearance, mental health and satisfaction with management. The first 22 items assess these domains and are scored on a scale of 1 (worst) to 5 (best). Depending on the version, additional items are scored on a variety of scales, for example on the SRS-30, item 23 asks about satisfaction with self-image, rated on a scale of 1 (very low) to 9 (extremely high), while items 24-30 ask about improvements post-surgery and are either rated on a scale of 1-3 or 1-5. Overall, studies reported generally high scores, indicating good HRQOL as measured by the SRS. These studies also made a number of comparisons, either between SRS scores and the characteristics of participants (i.e. curve progression or gender for example), type of intervention (the majority of which compared surgical interventions) and between different populations or groups, or a combination of these. The below discussion will focus on these comparisons.

Almost all studies that examined SRS results as they related to AIS and participant characteristics found little relation between SRS scores and those characteristics. A number of studies found no association between curve magnitude, postoperative correction rates, loss of correction (Benli et al., 2007), curve progression (Sudo et al., 2016), scoliosis classification (or curve type) or gender (Mueller & Gluch, 2009; Scaramuzzo et al., 2017). Of the studies that examined these results longitudinally, no significant difference in SRS scores were found to be associated with participant characteristics over time (Scaramuzzo et al., 2017).

A number of studies also compared SRS scores between different intervention types. There was only one study that reported on the impact of exercise. Lebel & Lebel (2016) reported that SRS-22 scores were consistently high (good) suggesting no changes in scores over the course of a Schroth physiotherapy intervention. Almost all other studies compared different surgical techniques. Three studies made no comparisons but suggested that SRS scores generally indicated good HRQOL amongst a number of samples who had undergone surgery (Kelly et al., 2010; Sudo et al., 2013a; Sudo et al., 2013b).

All other studies made comparisons either within or between surgical interventions. The majority of studies that examined surgical interventions also noted few differences in SRS scores between surgical groups: SRS scores were not associated with fusion level (Green et al., 2011) or between those who had Cotrell-Dubousset (CD) instrumentation still in situ opposed to those who had theirs removed because of complications (Mueller & Gluch, 2012). Only one study found difference between surgical groups. Remes et al. (2004) report that while almost all scores were similar, differences were found in follow up SRS-24 scores between those who had CD instrumentation and Universal Spine System (USS) instrumentation. A small but significant number of participants who had CD instrumentation reported pain often or very often, compared to nobody reporting pain often or very often in the USS instrumentation group. Furthermore, and like the above studies, characteristics of participants' AIS were not related to SRS-24 scores.

Several studies examined SRS data longitudinally adding a temporal element to these results. Similar to the studies above, SRS scores were not associated with fusion level (Lavelle et al., 2016) and remained largely steady before and after surgery and long-term post-surgery (Lavelle et al., 2016; Louer et al., 2019; Min et al., 2013). However, these studies did identify small numbers of participants who felt their function had worsened; Pérez-Grueso et al. (2000) for example, suggests that while the majority of participants were happy post-surgery, a small number were not, scoring lower on a number of SRS domains. In contrast, another study found significant differences across three points in time, suggesting that amongst a number of patients who had undergone surgery, SRS scores for pain improved at two years follow up, but were lost at ten years (Newton et al., 2020). Furthermore, improvements in self-image and total SRS scores from pre-operative scores, were maintained at 10 year follow up (Newton et al., 2020).

Beyond examining differences in (mostly) surgical techniques, a number of studies also looked more broadly at differences between groups - those who had AIS and had surgery, those who had brace treatment, those who went untreated and healthy controls, for example. These studies generally yielded a number of more significant results in comparison to those reported above. However, a small number of studies still found no differences in SRS scores between those having had selective thoracic fusion and age and gender matched controls (Enercan et al., 2015), those with CD instrumentation and controls (Pérez-Grueso et al., 2000) and those who had brace treatment or who were untreated (Danielsson et al., 2010). In

contrast a number of studies found significant differences in SRS scores between those who had a range of surgical interventions and controls (Kino et al., 2019), pregnant women who had previously had surgery and controls. Falick-Michaeli et al. (2015) suggested that women who underwent scoliosis correction surgery suffered from an increased incidence of long-term back pain after pregnancy compared to controls. Furthermore, SRS-24 scores suggested that six of the 17 of the women with AIS had severe back pain during pregnancy (35%) mandating home treatment or hospitalization, and 13 of the 17 women continued to experience sustained back pain that impacted their life after delivery of the child (76%). Newton et al. (2020) suggested that compared to previously published data of age-matched controls and AIS participants who had undergone a range of surgeries, at 10 year follow up both the male and female participants had more symptomatic pain compared controls. Furthermore, at 10-year follow-up, 23 patients (13%) had SRS-22 scores two standard deviations below the mean for normal individuals in the pain domain, in self-image, in function, and in mental health. However, in comparison with the subjects aged 20–40 years who had a spinal deformity but who were not surgically treated, participants who were surgically treated had substantially higher scores (better outcomes) in all domains. Finally, Erwin et al. (2020) suggest that pain and self-image were worse amongst those who had AIS, function was worse amongst females aged >40 with AIS and mental health scores for females with AIS were worse than males in AIS groups. This study goes on to report a number of other results, that touch upon all areas reported above, demonstrating a significant difference in scores across age groups, with older age groups scoring worse in all sub-scales of the SRS except mental health. Furthermore, among those with the most 'surgical-sized curves' those who had gone on to have surgery reported worse scores in function, pain, self-image and overall SRS-22 scores amongst those who had surgery.

*Oswestry Disability Index results*

Nine studies utilised the Oswestry Disability Index (ODI). The ODI is a tool that measures function related to lower back pain (Holm et al., 2003). The ODI has ten questions related to pain, personal care and activities of daily living. Each question is rated on a 6-point scale, from 0 (no pain) to 5 (worst imaginable pain). The sum of these scores is calculated and presented as a percentage, wherein 0% represents no pain and disability and 100% represents the worst pain and disability. Many of the results are similar to those found with the SRS above in that most scores generally indicated very good HRQOL, with somewhat mixed

results when comparisons were made. Also like the above studies, those which used the ODI also explored the relationship between ODI scores, characteristics of participants (or their AIS), differences between interventions or differences between populations or groups.

Studies that explored the relationship between ODI scores, intervention and participant characteristics report mixed results. There was no association between ODI scores and level of fusion (Green et al., 2011; Lavelle et al., 2016), level of instrumentation or with longer follow-up time (Lavelle et al., 2016). In contrast, Erwin et al. (2020) found that ODI scores positively correlated with age, BMI, and curve size, however (and similar to the SRS-22 results reported above) no relationship was found between ODI scores and participants age, or curve size (between surgical and non-surgical patients) although overall, ODI score was significantly worse in surgical patients.

Three studies found no significant relationship between ODI scores when compared to those with AIS and controls (Bjerkreim et al., 2007; Enercan et al., 2015; Kelly et al., 2010). In contrast, Kino et al. (2019) found that between those with CD instrumentation and controls, controls had significantly lower (better) ODI scores.

*Results of other scales*

One study utilised the Roland Morris disability questionnaire (RDQ). The RDQ is a 24-item self-report outcome measure that enquires about pain-related disability resulting from lower back pain (Roland & Morris, 1983). Items are scored 0 if left blank or 1 if endorsed, for a total RDQ score ranging from 0 to 24; higher scores represent higher levels of pain-related disability. Kino et al. (2019) reported a low overall mean (1.13) this was significantly higher than a control group. Kino et al. (2019) also utilised the Short Form – 36 (SF-36). The SF-36 consists of one item on recent changes in health and 35 items that are scored across eight subscales (Ware & Sherbourne, 1992). Lower scores indicate poorer health. Once again, scores were relatively high suggesting those with AIS were again, in generally good health; however, when compared to a population who did not have AIS, those with AIS had significantly lower scores in the physical and mental subscales (Kino et al., 2019).

Two further studies also used the SF-36. Danielsson et al. (2010) found that there was no difference between those with AIS who either had no treatment or treatment with a brace, but

when compared to a population of age matched controls, significant differences were found between the control group and non-treated AIS patients in physical functioning and general health subscales, and between brace treated patients and control group in physical functioning, 'role physical' (original author term), and social functioning subscales. Lavelle et al. (2016) found that SF-36 scores did not correlate with level of fusion or with those who had longer follow up.

Falick-Michaeli et al. (2015) utilised the Female Sexual Distress Scale – Revised (FSDS) (Derogatis et al., 2002) and the Beck Depression Inventory (BDI) (Beck et al., 1996).The FSDS is a 13-item scale that was created to assess sexually related distress. Each item is rated on a scale of 0 (never) to 4 (always), with scores summed to a maximum of 52, with higher scores indicating greater distress. The BDI has 21 items rated on a four- point scale (0-3), which is summed to maximum score of 63. The higher the score, the greater the symptoms of depression. Generally, scores between 14-19 indicate mild depression, 20-28 moderate depression and 29-63 severe depression. There were no significant differences in FSDS scores between two groups with AIS (those who were pregnant and those who were not) While mean scores on the BDI were lower amongst controls, again, there were no significant differences.

Bjerkreim et al. (2007) utilised the EuroQol (EQ-5D). The EQ-5D is a generic (non-disease specific) instrument developed to measure quality of life and quality-adjusted life-years (Balestroni & Bertolotti, 2012). The scale includes 5 items related to quality of life and a visual analogue scale for assessment of overall health. Amongst those with AIS, scores were noted to be slightly worse (however non-significant) when compared the general population. This study did note that 45% of participants had consulted a doctor or physiotherapist for back pain in the last 12 months before follow-up. Finally, one study included in this review utilised a researcher-developed questionnaire (Boos et al., 2007). The questionnaire covered the domains of back pain (4 items), function (3 items), self-image (3 items) and patient satisfaction (3 items), but how the items were scored was not described. Whilst approximately 50% of participants experienced no pain and did not require pain medication, the remainder indicated they had some degree of pain and required medication at least some of the time. In terms of daily function, no significant difference was found pre to post surgery. However, the majority of participant felt that AIS was at least somewhat of an influential factor in their career choice. Majority of participants also felt that surgery had

improved their appearance (64%), however the remainder felt that surgery had made no difference to their appearance. Most participants (76%) were also satisfied with their surgery.

## Discussion

Scoliosis Research Society scores in a majority of studies indicated that people with AIS at long-term follow up have generally good HRQOL. Almost all studies found little relationship between AIS or participant characteristics and SRS scores, that is, it was often found that SRS scores were relatively high regardless of the pre-operative curve magnitude or gender, for example. Between studies that explored different intervention types, again scores were high, however with the exception of one study there was little difference in SRS scores between different interventions, suggesting that there was no significant relationship between SRS scores and level of fusion or type of surgery. This generally held over time, again with most studies indicating little change in SRS scores at different points of follow up. Finally, amongst the studies that compared SRS scores between different populations, a clearer picture emerged. While three studies found no differences between surgical and brace treated AIS participants and controls, at least five studies reported that SRS results indicated that those with AIS had lower HRQOL compared to those who did not.

Oswestry Disability Index results painted a more mixed picture. With the exception of one study, it was clear that ODI scores had little relationship to participant or AIS characteristics. Two studies suggested that participants with AIS had lower HRQOL to healthy controls, however two other studies also suggested no difference.

Of the other instruments used, many produce similar results, generally indicating a good level of functioning and HRQOL amongst AIS patients. However, this level of function was often noted to be significantly lower than control or the general population, similar to a number of studies that utilised the SRS and ODI.

In summary, while there were some exceptions, much of the evidence suggests that HRQOL as measured by the above instruments is not significantly influenced by different AIS types, participant characteristics, surgery type or even over time. The evidence above does however begin to suggest that while AIS participants had a generally good quality of life, this is lower than those who do not have AIS.

There are some shortcomings related to the literature that has examined HRQOL in long-term outcomes of AIS that deserve discussion and may explain the seemingly disparate impression of outcomes between the studies reviewed here. HRQOL has been measured by only a handful of instruments. Although the SRS and ODI for example appear to be psychometrically robust in a number of ways, questions remain related to construct validity, that is, whether there is confidence that the above instruments thoroughly measure all elements that relate to HRQOL in this population. There are three likely explanations for why a number of studies did not find significant relationships between participant variables and HRQOL scales. First, there was no difference between the groups to begin with, so the ability to demonstrate change is limited. Second, the instruments used were not sensitive enough, that is, they failed to measure more subtle differences in HRQOL. Third, the instruments were not comprehensively measuring all aspects of HRQOL relevant to this population. We are sceptical of the first possibility and believe that the remaining two may be influenced by the manner in which the scales were designed. If there was no patient input, as in modern Patient Reported Outcome Measure (PROM) development (e.g., Rothrock et al., 2011), then there is substantial scope for further investigation into HRQOL with people diagnosed with AIS, for several reasons.

Firstly, the above studies stand in contrast to a rich literature that views HRQOL as a dynamic, relational and largely unsettled concept (Moons et al., 2006). There are various definitions and conceptualisations of HRQOL. The definition of HRQOL that is used in the development of a scale, the domains of HRQOL that are included in the scale, and the level of patient input into scale development will all impact the items and thus substantially affect the results obtained. We cannot and should not assume that the above scales discussed in this paper adequately reflect HRQOL aspects of concern to patients with AIS. . Secondly, the studies reviewed above stand in contrast to an emerging literature that has utilised qualitative techniques to explore the experiences of those with AIS. For example, Honeyman & Davison (2016) explored perioperative experiences of six participants with AIS. This was a stressful experience for both the patients and their parents, with the authors identify recurring themes related to the fear of the unknown, lack of control and more specific issues such as pain. Rullander et al. (2013) also found that surgery had a significant impact on all participants, who exhibited strong emotions before and long after surgery, with feelings such as fear, nightmares, nervousness, and helplessness emerging. Whilst there may have been a

participant selection bias (those with stronger negative emotional responses consenting to participate in the study) the findings point to the presence of these experiences amongst patients with AIS, and indicate that similar studies are needed to explore these issues and their impact on HRQoL in the longer term. Finally, anecdotal evidence gathered by the research team, either through publicly available social media platforms or personal contact with people with fused or unfused idiopathic scoliosis clearly indicates the profound impact the condition has on individuals' lives. There are personal reports from those living with fused and unfused AIS, of living permanently with chronic pain, a reliance on often multiple analgesics, and disrupted ability to work due to fatigue, pain, or discomfort.

There is a pressing need for qualitative research that explores the lived experiences of people with AIS. Such research would complement the studies reviewed here, allowing for a more rounded exploration of what HRQOL means for those with AIS and the factors that impact on HRQOL. This research would also inform future quantitative work and potentially lead to the development of other instruments to measure HRQOL in those with AIS, using modern methods which involve patients in instrument development. Additional work in these areas is likely to be extremely beneficial to patients with AIS, and their parents, who often make critical, life-long decisions about their health and wellbeing at an early age. Well-informed decision-making needs to be based on robust quantitative AND qualitative evidence, that reassures the patient and parents of the efficacy of surgery in addressing the curve, and the long-term outlook, in terms of quality of life.

## Conclusions

We have examined the long-term outcomes related to HRQOL for people with AIS, who had or had not had spinal fusion surgery. While there were some exceptions, most of the evidence suggests that HRQOL as measured by the above instruments doesn't appear to be significantly influenced between different AIS types, participant characteristics, surgery type or even over time. The evidence above does however begin to suggest that while AIS participants had a generally good quality of life, this is lower than amongst those who do not have AIS. Importantly, the available literature fails to address more fundamental questions about how HRQOL is conceptualised for those with AIS, and there is value in pursuing qualitative inquiry in this area.

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Table 1. Studies included in this review

| Author | Year | Aims | Country | Sample size | Methodology | Outcomes | Intervention/treatment | HRQOL meaures used |
|---|---|---|---|---|---|---|---|---|
| Benli et. al. | 2007 | This study examined surgical outcomes and Turkish SRS-22 questionnaire results of 109 late-onset adolescent idiopathic scoliosis patients surgically treated with third-generation instrumentation. | Turkey | n = 109 | Quantitative - Prospective cohort study | In terms of quality of life, this study essentially reports mean SRS-22 scores as low, moderate etc… | Surgical - Texas Scottish Rite Hospital System (n = 109) | The Scoliosis Research Society Patient Questionnaire (SRS-22). |
| Bjerkreim et. al. | 2007 | This study examined the long-term results after operative treatment with Cotrel-Dubousset (CD) instrumentation for adolescent idiopathic scoliosis (AIS). | Norway | n = 100 | Quantitative - Prospective cohort study | Radiologic results, patient satisfaction, and mean scores for quality of life and back function were excellent after CD instrumentation for AIS, but a considerable number of patients had treatment for back problems. | Surgical | EuroQol (EQ-5D) and ODI |
| Boos et. al. | 2007 | This study examined clinical and radiological results of Cotrel-Dubousset instrumentation (CDI) for the treatment of adolescent idiopathic scoliosis. | Swtizerland | n = 54 | Quantitative - Retrospective cohort study | In relation to HRQOL, overall patient satisfaction, functional status and subjective cosmetic improvement is high. | Surgical | Researcher developed questionnaire (13 items) measuring pain, function, self-image, patient satisfaction |
| Danielsson et. al. | 2010 | This study examined quality of life in adulthood in AIS patients receiving no active treatment, compared with similar patients who had been brace treated during adolescence. | Sweden | n = 77 | Quantitative - Case control study | The SRS-22/total score was a mean of 4.2 for braced patients and 4.1 for only observed patients. Neither total scores/subscales of the SRS-22 or SF-36 differed significantly between the groups. For the SF-36, no differences in relation to the Swedish age-matched norm scales were found for either group. Patients with moderate AIS report good quality of life in their 30s, as measured by both the SRS-22 and SF-36, regardless of whether they received no active treatment or were brace | Bracing - (n = 37); Observation - (n = 40) | SRS-22 and SF-36 |

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| | | | | | | treated during adolescence | | |
| Enercan et. al. | 2015 | This study evaluated the long-term behavior of the lumbar curve in patients with adolescent idiopathic scoliosis treated with selective thoracic fusion and to assess the clinical and radiologic outcomes in this fusion group compared with an age- and gender-matched group. | Turkey | n = 25 (AIS); n = 30 (controls) | Quantitative - Case control study | Our study group had excellent long-term clinical outcomes and showed no significant difference between patient and control groups in terms of all SRS domains. The mean ODI in the normal population was 3.7 , which is comparable with the mean 4.5 value in the AIS group. | Surgical - Selective thoracic fusion (n = 25). Controls - (n = 30) | SRS-22 and Oswestry Disability Index (ODI) |
| Erwin et. al. | 2020 | This study evaluated the impact of unoperated adolescent idiopathic scoliosis (AIS) in adulthood on pain, quality of life, and need for operative management. | US | n = 255 | Quantitative - Retrospective cohort study | Patients with AIS have SRS-22r scores that are lower than age–gender-matched controls in most domains. ODI had a positive linear correlation with age, body mass index, and curve size. | Observation - (n = 255) | SRS-22 and ODI |

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| Falick-Michaeli et. al. | 2015 | This study examined the rates of anesthesia prescription and satisfaction with surgery, prevalence and severity of low back pain, prevalence of depression, and sexual dysfunction among pregnant and nonpregnant patients with AIS undergoing correction surgery with pedicle-based systems and healthy woman with a history of pregnancy. | Israel | n = 40 (AIS); n = 6 (controls) | Quantitative - Retrospective cohort study | The SRS24 scores in the patients with AIS were 72% (88/120), showing a low score of 3.69/5 in the pain domains (p ¼ 0.0048 when compared with nonpregnant patients with AIS).<br>Depression rates were in the normal range and similar in all groups. FSDS scores, used to assess sexual dysfunction, were 4.02 in the pregnancy group and 5.67 in the nonpregnant<br>group (not significant) and 4.6 in the nonscoliosis control group (not significant). | Surgical - Pedicle screw (n = 40) | Female Sexual Distress Scale – Revised (FSDS), Beck Depression Assessment Questionnaire, SRS-24 |
| Green et. al. | 2011 | This study examined the long-term clinical and imaging results focusing on the ninstrumented lumbar spine after posterior spinal fusion for adolescent idiopathic scoliosis | US | n = 20 | Quantitative - Retrospective cohort study | SRS-22R scores and subscores were calculated demonstrating that all patients had good to excellent outcomes with an average overall score of 4.3 and average function and pain subscores of 4.5 and 4.3, respectively. Similarly, Oswestry<br>scores averaged 14.7 indicating minimal disability. | Surgical - Posterior fusion (n = 20) | SRS-22, ODI |
| Kelly et. al. | 2010 | This study evaluated a group of patients based on Scoliosis Research Society (SRS)-30 and Oswestry data as well as radiographic and magnetic resonance imaging (MRI) and report the results of long-term follow-upof this surgical treatment for this particular curve pattern in AIS. | US | n = 31 | Quantitative - Retrospective cohort study | The anterior approach in the treatment of thoracolumbar and lumbar curves in AIS offers good long-term functional outcomes for patients. Despite expected degenerative changes, patients scored well on the SRS and Oswestry tests, and were able to pursue careers and family activities. | Surgical - Anterior spinal fusion (n = 31) | SRS-30, ODI |

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| Kino et. al. | 2019 | This study investigated the long-term health-related quality of life (HRQOL), including marital and reproductive status, of middle-aged Japanese women who underwent posterior spinal fusion using CDI for AIS in its earliest days in Japan. | Japan | n = 29 (AIS); n = 71 (controls) | Quantitative - Retrospective cohort study | HRQOL scores in the patient group were generally lower than that in the healthy control group, although there was no significant difference between the two groups in the role component summary score (RCS) of SF-36. Marital and reproductive status were not significantly different between patient and control groups, and results for the patient group were similar to Japanese national data. | Surgical - CDI (n = 29) | Roland-Morris Disability Questionnaire, ODI, SRS-22, SF-36 |
| Lavelle et. al. | 2016 | This study examined long-term functional outcomes of patients treated with Cotrel-Dubousset (CD) instrumentation and determined whether distal level of instrumented fusion (L4 and L5) correlate with increased back pain or lower functional level. | US | n = 22 | Quantitative - Retrospective cohort study | No relationship was shown between ODI and distal level of fusion ($p = 0.72$). SF-36 and SRS 22 values were also not related to distal level of instrumentation. Patient reported VAS back pain scores ($r2 = 0.18$, $p = 0.05$), ODI ($r2 = 0.09$, $p = 0.17$), and SF-36 and SRS-22 were not worse in patients with longer follow-up over time. Back pain and certain functional score subcategories of the SF-36 and SRS-22 trended toward improved results over time. | Surgical - (n = 22) | SF-36, SRS-22, ODI, visual analog scale (pain) |
| Lebel & Lebel | 2016 | This case presentation shows that indeed adult curve progression can be stopped and even reversed with scoliosis specific Schroth physiotherapy (SSSPT) in an adult patient with scoliosis. | Canada | n = 1 | Quantitative - Retrospective case study | Within one month of beginning SSSPT, the patient reported no more back pain and within 2 months, reported improved breathing. The patient also benefitted from improved chest expansion, reduced scoliosis curve angles (measured in Cobb degrees), increased vital capacity, decreased ATR, and higher SRS-22 scores. She became more active and resumed all athletic activity within 8 months of beginning Schroth physiotherapy. | Exercise - scoliosis specific Schroth physiotherapy | SRS-22 |

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| Louer et. al. | 2019 | The purpose of this study was to report 10-year prospective radiographic and patient-rated outcomes of selective fusions of the main thoracic (MT) or thoracolumbar/lumbar (TL/L) curve, with particular attention to the behavior of the uninstrumented, compensatory curve. | US | n = 51 | Quantitative - Prospective cohort study | On the whole, SRS scores did not decrease during follow-up, and no patient had secondary operations. | Surgical - selective fusions (n = 51) | SRS-24 |
| Min, et. al. | 2013 | This study examined the clinical, radiological and pulmonary function results of 48 consecutive patients with 10-year minimal follow-up. | Switzerland | n = 48 | Quantitative - Prospective cohort study | SRS-24 scores did not change with time. There was no significant difference of 10-year SRS-24 scores between the those who had different levels of instrumented vertebra. patients who had the spinal implant removed for late infection also reported good subjective outcomes. | Surgical | SRS-24 |
| Mueller & Gluch | 2012 | This study explored long term outcomes among patients treated for AIS with CD instrumentation and spondylodesis. | Germany | n = 40 | Quantitative - Prospective cohort study | Only 14 out of 40 patients (35%) completed the SRS-24 questionnaire after a mean of 14.3 years postoperatively. In the SRS-24 questionnaire, the total score averaged 93.3 points out of a maximum 120 points (min. 71 to max. 106 points) at the follow-up. The analysis of the questionnaire showed no significant differences between the 5 patients with instrumentation still in situ (average 96.4 points) and the 9 patients after the removal of the instrumentation (average 91.5 points). | Surgical - Cotrel-dubousset instrumentation (n = 40). Only n = 14 completed SRS-24 | SRS-24 |

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| Mueller, et. al. | 2009 | This study examined long term outcomess in patients treated for AIS with a posterior titanium instrumentation. | Germany | n = 50 | Quantitative - Retrospective cohort study | SRS scores generally indicated that participatns had good HRQOL. Specifically, 48 of 49 patients (98%) were highly or fairly satisfied with the result of the treatment; only one female patient was somewhat dissatisfied – however we saw no objective (e.g. loss of correction) signs concerning this result. 42 of 49 patients (86%) reported to suffer never or rarely from back pain at rest. Overall 44 of 49 patients (90%) would definitely or probably undergo the same treatment again. | Surgical | SRS-24 |
| Newton et. al. | 2020 | This study examined the long term outcomes related to of spinal fusions for thoracic AIS: radiographic and clinical outcomes | US | n = 174 | Quantitative - Prospective cohort study | Spinal fusion patients report SRS-22 quality of life 10 years after scoliosis surgery that is minimally reduced compared to healthy peers and substantially better than an un-operated cohort of comparably aged scoliosis patients. Adolescents with thoracic idiopathic scoliosis should expect little if any change in their health-related quality of life compared to before surgery, high satisfaction, and a 7.5% chance of revision surgery 10 years after their index spinal fusion. | Surgical - Pedicle screw (n = 102), hook or hybrid constructs (n = 22), anterior screw rod constructs (n = 50) | SRS-22 |
| Perez-Grueso et. al. | 2000 | This study evaluated whether the use of instrumentation systems that preserve the sagittal profile could reduce the incidence of early degenerative changes | Spain | n = 34 (AIS); n = 35 (controls) | Quantitative - Retrospective cohort study | Cotrel–Dubousset instrumentation maintains the physiologic sagittal contour. Although there are some degenerative changes in magnetic resonance images and dynamic radiographs, the quality of life and daily activities of the patients after surgery are similar to those of a normal population of the same age. | Surgical - CDI (n = 35) | SRS *the authors refer to this as the SRS questionnaire - will need further information to determine what version it is. |

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| Remes et. al. | 2004 | This study examined whether there were any differences in outcome between the 2 instrumentation systems. | Finland | n = 112 | Quantitative - Case control study | The total SRS questionnaire score averaged 97 in the CD and 101 in the USS instrumentation groups at the final follow up. Six (11%) patients in the CD, but none in the USS instrumentation group, reported back pain often or very often on the SRS questionnaire | Surgical - CDI (n = 57), USS instrumentation (n = 55) | SRS |
| Scaramuzzo, et. al. | 2017 | This study examined the radiological and clinical outcomes of a single-center case series of selective thoracic fusions (STF) in adolescent idiopathic scoliosis. | Italy | n = 90 | Quantitative - Retrospective cohort study | Results suggest that all SRS 22 evaluations showed significant improvement at 6 months, in particular, the self-image analysis. A further improvement was also reported at 10 years, in particular in pain and function scores. | Surgical | SRS-22 |
| Sudo et. al. | 2013 | This study assessed the long-term outcomes of anterior spinal fusion (ASF) for treating thoracic adolescent idiopathic scoliosis (AIS). | Japan | n = 25 | Quantitative - retrospective cohort study | The average total SRS-30 score was 4.0. Overall radiographical findings and patient outcome measures of ASF for Lenke 1 MT AIS were satisfactory at an average follow-up of 15 years. | Surgical - Anterior spinal fusion (n = 25) | SRS-30 |
| Sudo et. al. | 2013 | This study examined the outcomes of anterior dual-rod instrumentation in a consecutive series of patients with thoracolumbar/lumbar adolescent idiopathic scoliosis managed by a single surgeon at a single institution. | Japan | n = 30 | Quantitative - Retrospective cohort study | The average total SRS-30 score was 4.2. Radiographic findings, pulmonary function, and clinical measures were satisfactory at the time of followup, at a minimum of twelve years. | Surgical - Dual -rod instrumentation (n= 30) | SRS-30 |
| Sudo, et. al. | 2016 | This study examined long-term outcomes about the behaviour of the thoracolumbar/lumbar (TL/L) curve after thoracic anterior correction and fusion (ASF) and to determine the impact of ASF on pulmonary function | Japan | n = 14 | Quantitative - Retrospective cohort study | Results suggest that the mean SRS-22 total score at the final follow-up was 4.0 (SD 0.3). No significant difference was observed between patients with and without a TL/L curve increase | Surgical | SRS-22 |

Table 2. Summary of outcome measures used and results

| Instrument | How this instrument measures HRQOL | Studies | Significant or notable results |
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| Scoliosis Research Society Questionnaire (SRS) | The SRS has a number of variants (SRS-22, 23, 24 and 30) While items differ slightly across versions, the SRS contains questions that examine five domains: function/activity, pain, self-image/appearance, mental health and satisfaction with management. The first 22 items assess these domains and are scored on a scale of 1 (worst) to 5 (best) with a higher score indicating better HRQOL. | n = 21: (Benli et al., 2007; Danielsson et al., 2010; Enercan et al., 2015; Erwin et al., 2020; Falick-Michaeli et al., 2015; Kelly et al., 2010; Kino et al., 2019; Lavelle et al., 2016; Lebel & Lebel, 2016; Louer et al., 2019; Min et al., 2013; Mueller & Gluch, 2009, 2012; Newton et al., 2020; Pérez-Grueso et al., 2000; Remes et al., 2004; Scaramuzzo et al., 2017; Sudo et al., 2013a; Sudo et al., 2013b; Sudo et al., 2016) | Overall studies suggested that mean SRS scores were high (good). No differences were found between participant characteristics (i.e. curve type and gender) and SRS scores. Few differences were found between surgical and non-surgical interventions. SRS scores were generally stable over time. A number of studies noted differences in SRS scores between those with AIS and controls, with controls generally reporting higher (better) SRS scores. |
| Oswestry Disability Index (ODI) | The ODI is a tool that measures function related to lower back pain. The ODI has ten questions related to pain, personal care and activities of daily living. Each question is rated on a 6-point scale, from 0 (no pain) to 5 (worst imaginable pain). The sum of these scores is calculated and presented as a percentage, wherein 0% represents no pain and disability and 100% represents the worst pain and disability. | n = 7: (Bjerkreim et al., 2007; Enercan et al., 2015; Erwin et al., 2020; Green et al., 2011; Kelly et al., 2010; Kino et al., 2019; Lavelle et al., 2016) | Overall, studies reported low (good ODI) scores, however results were somewhat mixed. Two studies indicated no relationship between ODI score and level of fusion, however one study found that ODI scores were positively correlated with age, BMI and curve size. Two studies found no difference between those with AIS and controls, while one study suggested that controls had significant lower (better) ODI scores. |
| Short Form 36 (SF-36) | The SF-36 consists of one item on recent changes in health and 35 items that are scored across eight subscales. Lower scores indicate poorer health. | n = 3: (Danielsson et al., 2010; Kino et al., 2019; Lavelle et al., 2016) | Overall, SF-36 scores were relatively high (good). One study indicated that SF-36 scores were not related to level of fusion. However SF-36 scores were lower amongst those with AIS and controls in two studies. |
| Roland Morris Disability Questionnaire (RDQ) | The RDQ is a 24-item patient-reported outcome measure that enquires about pain-related disability resulting from lower back pain. Items are scored 0 if left blank or 1 if endorsed, for a total RDQ score ranging from 0 to 24; higher scores represent higher levels of pain-related disability | n = 1: (Kino et al., 2019) | RDQ scores were higher (worse) amongst those with AIS when compared with controls. |
| Female Sexual Distress Scale (FSDS) | The FSDS is a 13-item scale that was created to assess sexually related distress. Each item is rated on a scale of 0 (never) to 4 (always), with scores summed to a maximum of 52, with higher scores indicating greater distress. | n = 1 (Falick-Michaeli et al., 2015) | There were no significant differences between two groups with AIS (those who were pregnant and those who were not) and controls in regards to FSDS scores. |
| Beck Depression Inventory (BDI) | The BDI has 21 items rated on a four- point scale (0-3), which is summed to maximum score of 63. The higher the score, the greater the symptoms of depression. | n = 1 (Falick-Michaeli et al., 2015) | There were no significant differences between two groups with AIS (those who were pregnant and those who were not) and controls in regards to BDI scores. |
| EuroQol (EQ-5D) | he EQ-5D is a generic (non-disease specific) instrument developed to measure quality of life and quality-adjusted life-years. The scale includes 5 items related to quality of life and a visual analogue scale for assessment of overall health. | n = 1 (Bjerkreim et al., 2007) | While EQ-5D scores were elevated amongst those with AIS when compared to the general population, this was not significant. |
| Researcher developed | The questionnaire covered the domains of back | n = 1: (Boos et al., 2007) | These results suggest that 50% of participants experienced no pain |

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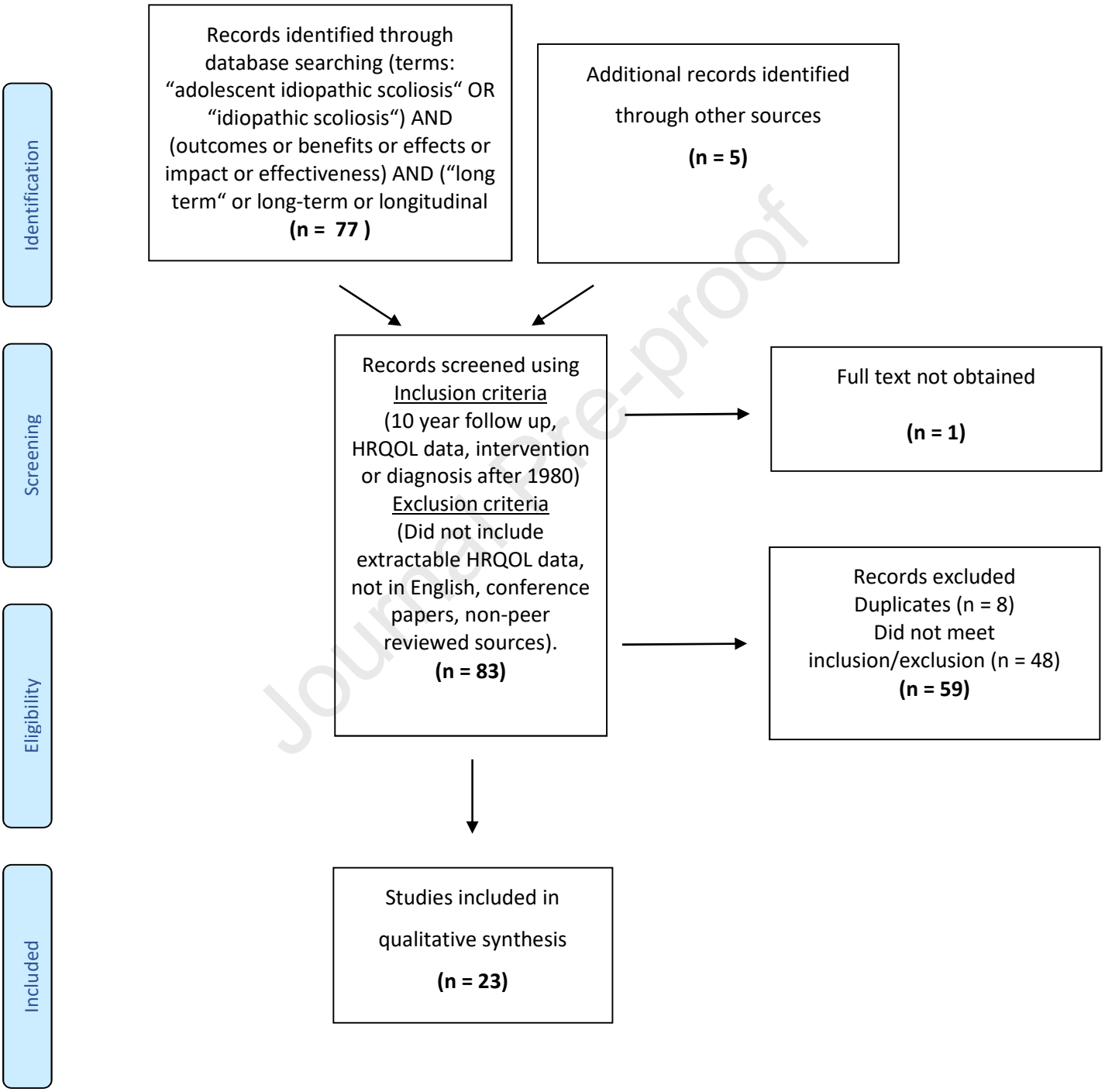
| scales | pain (4 items), function (3 items), self-image (3 items) and patient satisfaction (3 items), but how the items were scored was not described. No information on scoring provided | | and did not require pain medication, the remainder indicated they had some degree of pain and required medication at least some of the time. No significant differences were found in daily function pre and post surgery. The majority of participants indicated that AIS was an influential factor in their career choice. Majority of participants also felt that surgery had improved their appearance (64%), and were satisfied with their surgery (76%). |

Figure 1. PRISMA 2009 Flow Diagram (Moher et al., 2009)



This is a review paper, therefore ethical approval was not sought. We were mindful of ethical publishing practices when composing this manuscript and believe it is consistent with all major ethical standards.

Authors' contributions

LD, PN and MD identified the need for this review. RE and LD screened the identified papers for inclusion and conducted the quality appraisal. RE conducted the literature search, data synthesis and drafted the initial manuscript. GB and MD provided practitioner and patient input. All authors contributed equally to subsequent manuscript revisions.

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Conflicts of interest/Competing interests

LD and MD are mother and daughter. This review came about at the request of MD, who having undergone long thoracolumbar spinal fusion at aged 13, asked LD - in her capacity as a qualitative researcher – to undertake qualitative research into long-term psychosocial and socio-economic consequences of fusion surgery, because of the lack of current evidence. This literature review is the first in a series of projects aiming to address that request.