



# Long-term follow-up of untreated Scheuermann's kyphosis

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Received: 1 December 2019 / Accepted: 12 April 2021 / Published online: 1 July 2021  
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## Abstract

**Study design** Long-term cross-sectional study.

**Objectives** To investigate the long-term effects of untreated Scheuermann's kyphosis on quality of life, and its relationship to radiographic parameters of spinal deformity.

**Summary of background data** Previous studies reported reduced self-image, increased pain and impaired physical status. Little is known of the long-term impact of sagittal plane deformity in untreated SK.

**Methods** One hundred and thirteen consecutive untreated patients with SK were identified from a national service database prior to 2000, when surgery was not offered at this unit. 81 of these patients were available for evaluation; 66 (81%) consented to questionnaire and clinical evaluation, and 47 (58%) consented to additional radiological evaluation. Health-related quality of life (HRQoL) was compared to normative population values. Mean age was 45.1 years (31–65), and mean follow-up was 27 years (16–36). 57 patients had thoracic kyphosis and 9 had thoracolumbar deformity.

**Results** SRS-22 and SF-36 scores were lower, and ODI was greater in patients with untreated SK compared to normative population values. Kyphosis progressed from mean 66° at skeletal maturity to 78° ( $p < 0.001$ ) after mean follow-up of 27 years. Long-term progression of untreated SK was 0.45°/year ( $n = 47$ ). Multilinear regression showed good correlation between increasing SVA and worse ODI scores ( $r = 0.59$ ;  $p = 0.001$ ). Increasing SVA also correlated with worse function, pain and mental health scores reported by SRS-22, and with worse physical function and bodily pain scores reported by SF-36. Increasing CL correlated with worse SF-36 physical function scores. Increasing cSVA and increasing TK correlated with worse SRS-22 self-image scores.

**Conclusion** SRS-22 and SF-36 scores were lower, and ODI was greater in patients with untreated SK compared to normative data. Long-term progression of untreated SK was 0.45°/year ( $n = 47$ ). Increasing SVA correlated with worse SF-36 physical function, SRS-22 function, SRS-22 pain and higher ODI scores. Total kyphosis (TK) and cSVA were independent predictors of low SRS self-image.

**Level of evidence** III.

**Keywords** Scheuermann's · Kyphosis · Disease · Natural history · Outcome

## Introduction

Excessive kyphosis in youngsters was initially described by Hagelund [1]. Scheuermann described the radiographic features of juvenile kyphosis in 1920, comprising vertebral body wedging and endplate narrowing [2]. The cause of Scheuermann's kyphosis remains unknown; there is likely a strong, but as yet uncharacterised, genetic predisposition to the disease [3]. Disorganised endochondral ossification has been demonstrated in patients with Scheuermann's kyphosis, and mechanical factors are likely to contribute to the development of the increased kyphosis [4, 5].

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Scheuermann's kyphosis is diagnosed radiographically using Sorenson's criteria [6]. The most accurate population prevalence estimates from a prospective Dutch cohort study indicate the prevalence of Scheuermann's kyphosis as 4.0%; the prevalence amongst males was 4.5%, and females 3.6% [7]. The prognosis for patients with untreated Scheuermann's kyphosis remains unclear. The purpose of this study was to establish the natural history for patients with untreated SK using HRQoL measures, and to investigate the relationship of these with radiographic sagittal parameters of spinal deformity.

## Patients and methods

One hundred and thirteen consecutive untreated patients with Scheuermann's kyphosis were identified from the database of our national spine deformity service prior to 2000, when surgical treatment for the condition was not offered at this unit. Patients were identified on the Community Health Index, which is a register of all patients in the National Health Service in Scotland. SK was defined as contiguous wedging of three vertebral bodies at the apex of the kyphosis with typical endplate irregularity. Eighty-one patients were available for evaluation; six patients had died, six patients did not have capacity to consent, and twenty were no longer resident in Scotland. Clinical health care records were available for all patients. All patients were sent a recruitment letter, information sheet and written consent form, to which they responded if they wished to participate. Patients were given the option to participate in a questionnaire study, as well as additional clinical and radiographic evaluation. Of the 81 participants, 66 (81%; 39 males, 27 females) consented to clinical review only and 47 (58%) consented to additional radiological evaluation. Six patients received brace therapy until skeletal maturity. Three patients had undergone lower back surgery during follow-up; two patients underwent single-level lumbar discectomy for symptomatic disc herniation, and one patient underwent lumbar decompression for central canal stenosis. Values for initial radiographic measurements of participants were performed at skeletal maturity, at a mean age of 18.1 years (range: 15.1–34.4). The mean age of participants at follow-up was 45.1 years (range: 31–65), resulting in a mean follow-up of 27 years (range: 16–36). There were 57 patients (86%) who had thoracic SK, and 9 patients (14%) had thoracolumbar SK with an apex at T10 or lower. Ethical approval for this study was provided by NHS Lothian Research Ethics Committee (NHS Lothian REC No: 14/SS/1040).

## Long-term outcome assessment

Whole spine lateral radiographs were performed in a natural standing position, looking horizontally, hands resting on a vertical support [8]. Radiographic measurements were performed prospectively at initial presentation and at long-term follow-up. Radiographs were measured with the aid of software (Surgimap Version 2.2.9.9.8 for Windows, NY) for the following sagittal parameters: C2–C7 lordosis (CL), T1-slope, C2–C7 sagittal vertical axis (cSVA), T1 spinopelvic inclination (T1SI), pelvic incidence (PI), pelvic tilt (PT), sacral slope (SS), C7 SVA (SVA), T4–T12 kyphosis (T4–T12TK), total thoracic kyphosis (TK), L1–S1 lordosis (LL), total lumbar lordosis (TLL) and T11–L1 kyphosis (TLK) [9, 10].

Health-reported quality of life measures (HRQoL) were compared to population-, age- and sex-matched normative values. The Medical Outcomes Study 36-item short-form health survey (SF-36) was used to provide an overview of the participants' general health status, using normative data from the UK population for comparison [11, 12]. The Oswestry Disability Index (ODI; version 2.0) was used to assess pain level and impact of the disease on activities of daily living [13]. The disease-specific SRS-22 questionnaire was used to assess participants' self-image and appearance [14, 15]. For the SF-36 questionnaire, each domain is transformed into a 0–100 scale; lower scores indicate greater disability, and higher scores indicate less disability. The ODI overall score increases with increasing disability; 0–20% represents minimal disability, and 81–100% represents the most severe disability. The scores of each SRS-22 domain (function, pain, self-image and mental health) decrease with more severe symptoms. For the SRS-22 questionnaire, severe symptoms are represented by a lower mean domain score, and minimal symptoms are represented by a higher mean domain score.

## Statistical analysis

Correlation analysis of ODI, SF-36, SRS-22 scores and sagittal parameters were performed using Pearson correlation. Collinearity diagnostics, normality testing of datasets and multiple regression analysis were performed (SPSS v23.0). Comparison of means across multiple groups was performed by ANOVA with post hoc Tukey tests for comparison of means between subsequent individual groups. Statistical significance was accepted at  $p < 0.05$ , unless stated otherwise.

## Results

### Health-reported quality of life measures (HRQoLs)

HRQoLs were compared between untreated patients with SK and normative values for the population (Table 1). All subdomains of the SRS-22 and SF-36 were substantially lower, and ODI was substantially higher, in untreated SK patients compared to the normative population values. The body diagram of the SRS-22 questionnaire was analysed. Patients reported thoracic and shoulder pain in 66%, thoracolumbar in 49% and low back pain in 29%; leg pain was reported in 17%, and arm pain in 8%. No pain was reported by 18% of participants.

### Sagittal radiographic measurements

The mean initial TK of participants at skeletal maturity was 66.0° (50–100) and had progressed over follow-up (mean 27 years) to 78.2° (50–110;  $p=0.00001$ ). The mean initial TK for non-participants was 69.2° (48–90) and for deceased patients was 78° (55–110). There was no significant difference in the mean initial TK between participants, non-participants and deceased patients ( $p=0.232$ ). The mean rate of progression of TK amongst participants was 0.45°/year ( $n=47$ ); TK increased in 38 patients (80.9%; mean follow-up 30 years) and did not increase in nine patients (19.1%; mean follow-up 27 years) across the study period. The rate of TK progression did not depend upon age ( $p=0.07$ ),

gender ( $p=0.358$ ) or severity of initial kyphosis ( $p=0.05$ ). The mean rate of progression of TLK amongst participants was 0.58°/year. There was no significant difference in the rate of progression between participants with TSK and participants with TLSK ( $p=0.8$ ). There were no significant differences in ODI, SF-36, SRS-22 scores or subdomains between TSK and TLSK patients. TLK, LL and spinopelvic parameters did not correlate with any ODI, SF-36, SRS-22 or any HRQoL subdomains. Figure 1 shows radiographs and HRQoL outcomes for a patient with progression of thoracic Scheuermann’s kyphosis.

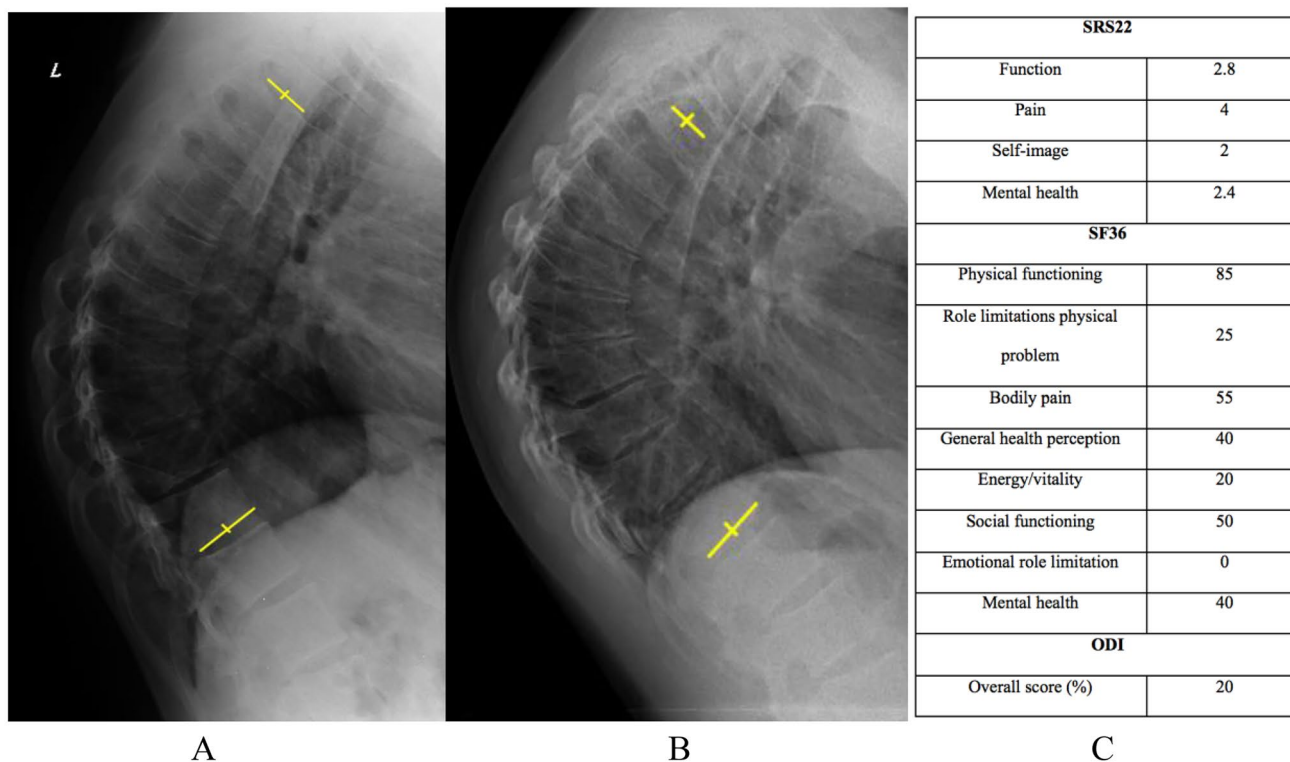
Sagittal parameters at final follow-up for the whole cohort of patients available for radiographic evaluation are shown in Table 2. Comparison of radiographic deformity characteristics between patients with TSK or TLSK is also shown in Table 2; the only significant difference between patients with TSK and TLSK was that patients with TLSK had significantly greater thoracolumbar kyphosis.

Age correlated weakly with TK ( $r=0.33$ ,  $p<0.05$ ) and SVA ( $r=0.36$ ,  $p<0.05$ ). Correlation between sagittal parameters was also performed. SVA correlated very strongly with T1-inclination ( $p=0.0001$ ;  $r=0.813$ ). There was a wide range in SVA values across the whole cohort (– 75 to 99, interquartile range: – 19.5 to 28). SVA correlated weakly with TK ( $r=0.292$ ;  $p=0.04$ ), CL ( $r=0.381$ ;  $p=0.01$ ), cSVA ( $r=0.331$ ;  $p=0.03$ ) and LL ( $r=0.321$ ;  $p=0.03$ ). T1 slope showed strong correlation with TK ( $r=0.601$ ,  $p=0.06^{-4}$ ). There was a very strong correlation between T4-T12 and total kyphosis ( $r=0.910$ ,  $p=0.01^{-19}$ ). Thoracolumbar kyphosis correlated negatively with CL

**Table 1** Comparison of HRQOLS between untreated SK patients and population normative values; the domains and values of components of the SRS-22 questionnaire are displayed against values obtained from control individuals

	Untreated SK patients [ $n=66$ ; mean (SEM)]	Population normative values (mean)
	SRS22	
Function	3.4 (0.1)	4.3
Pain	3.4 (0.2)	4.4
Self-image	2.8 (0.1)	4.2
Mental health	3.3 (0.1)	4.2
	SF36	
Physical functioning (PF)	68.8 (4.0)	89.6
Role limitations physical problem (RL)	65.0 (5.6)	85.2
Bodily pain (BP)	58.0 (3.8)	82.9
General health perception (GHP)	52.4 (3.4)	73.5
Energy/vitality (EV)	47.4 (3.0)	64.8
Social functioning (SF)	67.8 (3.8)	89.4
Emotional role limitation (ERL)	59.8 (5.5)	88.4
Mental health (MH)	61.5 (3.2)	76.5
	ODI	
Overall score	22.1 (2.8)	10.2

SK Scheuermann kyphosis, SF short form, SRS Scoliosis Research society, ODI Oswestry Disability Index



**Fig. 1** Example of a patient with progression of thoracic Scheuermann’s kyphosis: **a** lateral standing radiograph of male patient aged 31 years with thoracic kyphosis (T4–T12) measuring 82-degrees; **b** lateral standing radiograph of same male patient at 10-year follow-up

(aged 41 years), with thoracic kyphosis measuring 91-degrees. The outcomes of HRQoLs reported by this patient at follow-up (aged 41 years) are shown (c)

**Table 2** Sagittal radiographic parameters at final follow-up for the whole cohort of SK patients, patients with thoracic SK (TSK) and patients with thoracolumbar SK (TL SK); radiographic measures for

the reported parameters are shown as mean (range) for the patients available for radiographic evaluation ( $n=47$ )

Sagittal parameter	Whole cohort (SK, $n=47$ )	Thoracic SK (TSK, $n=42$ )	Thoracolumbar SK (TL SK; $n=5$ )
Cervical lordosis (C2–C7; CL,°)	23.1 (– 13 to 51)	24 (2–51)	15.8 (– 13 to 40)
Sagittal vertical axis (SVA; mm)	4.8 (– 75 to 99)	8 (– 44 to 99)	–19.8 (– 75 to 53)
Cervical SVA (cSVA; mm)	34.1 (11–75)	35 (11–75)	26.8 (21–37)
T1-slope (°)	38.4 (8–65)	39.4 (20–65)	30.2 (8–43)
Thoracic kyphosis (total kyphosis; K,°)	78.2 (50–110)	78.5 (53–110)	75.6 (50–90)
Thoracic kyphosis (T4–T12; TK,°)	71.4 (50–110)	71.8 (53–110)	67.6 (50–93)
Thoracolumbar kyphosis (T11–L1; TLK,°)	12.9 (– 10 to 43)	10.9* (– 10 to 34)	30.2* (23–43)
Lumbar lordosis (L1–S1; LL,°)	69.6 (43–96)	68.1 (43–92)	82 (65–96)

\*There were no significant differences between sagittal parameters between patients with thoracic SK compared with thoracolumbar SK, except for thoracolumbar kyphosis ( $p=0.0003$ )

( $r= - 0.342, p=0.02$ ). Lumbar lordosis correlated moderately with TK ( $r=0.410, p=0.03$ ). Therefore, SVA, cSVA, CL and TK were used for subsequent stepwise multiple regression analysis to identify significant relationships between sagittal parameters and subdomains of HRQOL measures and questionnaires.

Multilinear regression showed good correlation between increasing SVA and worse ODI scores indicating increasing SVA correlated with increasing disability ( $r=0.59; p=0.001$ ). It is important to understand that more severe symptoms and greater disability are reflected by low SRS-22 scores, low SF-36 scores and high ODI scores. Increasing

SVA also correlated with worse function ( $r = -0.52$ ;  $p = 0.004$ ), pain ( $r = -0.56$ ;  $p = 0.01$ ) and mental health scores ( $r = -0.47$ ;  $p = 0.03$ ) reported using the SRS-22. Increasing SVA also correlated with worse physical function ( $r = -0.54$ ;  $p = 0.003$ ) and bodily pain scores ( $r = -0.53$ ;  $p = 0.002$ ) reported using SF-36. Increasing CL also correlated with worse SF-36 physical function scores ( $r = -0.48$ ;  $p = 0.03$ ). Increasing cSVA ( $r = -0.44$ ;  $p = 0.03$ ) and increasing TK ( $r = -0.55$ ;  $p = 0.02$ ) correlated with worse SRS-22 self-image scores (Table 3).

## Discussion

There is limited evidence detailing the long-term patient-reported outcomes and prognosis of untreated patients with SK. Our study reported HRQoLs at long-term follow-up in a large cohort of untreated patients with severe SK. This study also assessed progression of deformity in these patients, reporting sagittal radiographic parameters and their correlation with validated HRQoLs.

Four previous studies have reported the natural history of untreated SK [16–19]. Murray et al identified a mean kyphosis of  $71^\circ$  in 67 patients over 32 years' follow-up [16]. Ristolainen reported patient outcomes in 80 SK patients (mean initial TK of  $45^\circ$ ) over 37 years' follow-up [17]. Damborg reported the general health status of 943 patients with SK identified from the Danish Twin Registry [18]. Ristolainen also reported radiographic progression of thoracic kyphosis in 19 patients over 46 years' follow-up [19]. To our knowledge, our study is the first to report both disease-specific HRQoLs and radiographic disease progression in a cohort of patients with severe untreated SK.

The quality of life in patients with untreated severe SK is incompletely understood. The most comprehensive study

reporting the long-term prognosis of untreated SK was that performed by Murray, which compared SK patients' questionnaire results with matched controls [16]. Patients with SK worked in lighter jobs than did control individuals, had more severe back pain, but had little preoccupation about their appearance, did not appear to be disabled by their symptoms, and had no significant limitation in activities of daily living. Damborg reported the general health status of patients of the Danish Twin Registry with self-reported SK. In this questionnaire study patients reported slight, but statistically inferior, differences in the physical component score of the SF-12 questionnaire [18]. Ristolainen reported that SK patients had a 2.5-fold increased risk of constant pain, an increased risk of back-pain related disability and sciatic pain after mean follow-up of 37 years, compared to control individuals. SK patients also reported statistically significant lower quality of life and lower general health status than control individuals. In comparison with other musculoskeletal conditions, we identified that patients with untreated severe SK reported SF-36 bodily pain and physical function scores similar to osteoarthritis of the hip and knee [20].

The prevalence of severe thoracic back pain has been reported in 50% of adult SK patients [21]. Sorenson reported thoracic pain in 50% of adolescent patients and 25% of adult patients [6]. Patients with SK complain of back pain and pain of greater intensity more frequently than control individuals [16, 17]. We identified that SK patients most frequently reported thoracic and shoulder pain, followed by thoracolumbar and lower back pain.

There is no longitudinal study describing changes in thoracic kyphosis with age in a normal population. Cross-sectional studies have estimated thoracic kyphosis to progress by  $3.6^\circ$  in males, and  $5.9^\circ$  in females from the third to fifth decades of life in North Americans ( $26.3^\circ$ – $29.8^\circ$ , and  $26.8^\circ$ – $32.7^\circ$ , respectively [22]), and by  $1^\circ$  in males and

**Table 3** Regression analysis between HRQoLs and sagittal radiographic measurements; individual domains of HRQoLs are shown in the column sub-headings

Sagittal parameter	HRQoL domain							
	SRS: function	SRS: pain	SRS: Self-image	SRS: mental health	ODI	SF-36: physical function	SF-36: bodily pain	SF-36: mental health
SVA	<b>(<math>r = -0.52</math>; <math>p = 0.004</math>)</b>	<b>(<math>r = -0.56</math>; <math>p = 0.01</math>)</b>	( $p = 0.96$ )	<b>(<math>r = -0.47</math>; <math>p = 0.03</math>)</b>	<b>(<math>r = 0.59</math>; <math>p = 0.001</math>)</b>	<b>(<math>r = -0.54</math>; <math>p = 0.003</math>)</b>	<b>(<math>r = -0.53</math>; <math>p = 0.002</math>)</b>	( $p = 0.31$ )
cSVA	( $p = 0.72$ )	( $p = 0.92$ )	<b>(<math>r = -0.44</math>; <math>p = 0.03</math>)</b>	( $p = 0.08$ )	( $p = 0.7$ )	( $p = 0.91$ )	( $p = 0.40$ )	( $p = 0.17$ )
CL	( $p = 0.42$ )	( $p = 0.05$ )	( $p = 0.06$ )	( $p = 0.36$ )	( $p = 0.2$ )	<b>(<math>r = -0.48</math>; <math>p = 0.03</math>)</b>	( $p = 0.12$ )	( $p = 0.68$ )
TK	( $p = 0.92$ )	( $p = 0.44$ )	<b>(<math>r = -0.55</math>; <math>p = 0.02</math>)</b>	( $p = 0.9$ )	( $p = 0.78$ )	( $p = 0.68$ )	( $p = 0.26$ )	( $p = 0.78$ )

The Pearson correlation factor ( $r$ ) for each sagittal parameter with each HRQoL sub-domain is shown. The significance ( $p$  value) is shown for correlation of each sagittal parameter with HRQoLs. For correlations with significant  $p$  values ( $p < 0.05$ ; shown in bold), the value of the associated Pearson correlation factor ( $r$ ) is also shown. Pearson correlation factors are not shown for non-significant correlations

2° in females from the third to fifth decades of life in Japanese (34.9°–35.9°, and 33.9°–35.9°, respectively [23]). Over 27 years' follow-up in our study, we observed progression of TK in 86.4% of untreated SK patients. The mean rate of TK progression was 0.45°/year in the whole cohort. There was no difference in progression of TK between genders. The mean rate of TK progression in our cohort was higher, but similar, to that reported by Ristolainen (0.30°/year); their cohort included patients with a lower baseline TK of 46°. It is unknown whether or not patients with larger TK have a greater rate of progression.

Our cohort demonstrated an increase in SVA and TK with age. A cross-sectional study in the Japanese normal population reported an increase in these parameters with a comparable mean SVA at a mean age of 45 years (2.8 mm vs 4.8 mm) [23]. In our cohort, SVA correlated most strongly with CL, followed by reduced lumbar lordosis, increasing cSVA and increasing TK. We identified that an increased TK was associated with an increased CL, while patients with a large TLK had less CL. This is similar to previous studies that also described increased TK leading to an anteriorly projected cervical spine, whereas TLK was associated with a vertically oriented cervical spine [24]. Increased TK is associated with increased cSVA and CL, which may contribute to increased pain [25].

A significant correlation has been reported between increasing kyphotic curve magnitude and lower scores, indicating more severe symptoms, for each SRS-24 sub-domain [26]. Interestingly, deteriorating self-image had the strongest correlation with increasing kyphosis. In Murray's study, patients demonstrated an increasing concern for self-appearance as the magnitude of the kyphosis increased, though the concern lessened with age [16]. In our cohort, we observed a moderate correlation between increasing TK and worse SRS-22 self-image scores ( $r = -0.55$ , Table 3). The mean SRS self-image value in our untreated severe SK patient cohort was similar to that reported in pre-operative adolescent SK patients [26]. Patients with SK often have forward protrusion of the head (described as a gooseneck deformity); that cSVA was an independent predictor of worse SRS-22 self-image scores in our study indicates that patients with SK are aware of this forward head protrusion, which negatively affects their self-image (Table 3).

Unoperated adult patients with spinal deformity and positive sagittal balance (SVA) have significant compromise in reported health and quality of life, as measured by SF-12, pain, function and self-image and SRS-22 domains [27]. Consistent with previous reports, our cohort of untreated SK patients demonstrated a significant correlation between increasing SVA and increased pain and disability, as demonstrated by worse ODI, SRS-22 pain, SRS-22 function and SF36 physical function scores.

Our study has strengths and limitations. It is a longitudinal cohort study of untreated patients with severe SK with a high rate of long-term participation, and radiographic follow-up. We utilised validated and disease-specific questionnaires to determine the health status of a cohort of patients with severe SK that remained untreated before surgical treatment was recommended by our service. Although all patients in this cohort were managed non-operatively, our study is at risk of selection bias as it is possible that patients with symptoms may be more eager to attend for clinical review and radiographic evaluation. We acknowledge that analysis of HRQoLs used normative population values for comparison and not age- and sex-matched controls, which would have been ideal, but are not available.

Clinical correlation of radiographic parameters is difficult. We reported radiographic sagittal parameters in untreated patients with SK. We reported regional sagittal parameters and SVA as a global sagittal parameter. Further global sagittal parameters such as T1 pelvic angle (T1PA) and global sagittal angle (GSA) may be helpful to assess global sagittal profile, though these were not measured in this study [28]. Larger numbers of patients completing clinical, questionnaire and radiographic evaluation would have been desirable. Due to the low proportion of patients with TLSK, our results should be interpreted with caution in this subgroup of patients with SK.

## Conclusion

Our study assessed HRQoL measures and their relationship to progression of deformity in a longitudinal cohort of untreated patients with Scheuermann's kyphosis. The extent of disability was greater in patients with untreated SK, compared to the normal population, as indicated by results in our cohort of untreated SK patients who reported lower SRS-22 and SF-36 scores, and greater ODI scores compared to normative population values. The long-term rate of progression of untreated SK was 0.45°/year. Increasing SVA correlated with a decreasing SF-36 physical function, SRS-22 function, SRS-22 pain and higher ODI scores. Total kyphosis (TK) and cSVA were independent predictors of low SRS-22 self-image.

## Key points

- Subjective quality of life measures are worse in patients with untreated Scheuermann's kyphosis compared to the normative population values.
- Long-term progression of untreated Scheuermann's kyphosis is 0.45°/year.

- Total kyphosis and cSVA were independent predictors of low reported self-image.
- SVA correlated with increased disability and severity of symptoms, as reported by health-related quality of life (HRQoL) measures.

**Acknowledgements** The authors would like to acknowledge the help of Richard A Parker (Senior Statistician, Edinburgh Clinical Trials Unit, University of Edinburgh) in the statistical analysis of results.

**Author contributions** EG: study design, data acquisition, data analysis and interpretation, drafting manuscript and final approval of manuscript. SR: data analysis and interpretation, drafting manuscript and final approval of manuscript. AD: data analysis and interpretation, drafting manuscript and final approval of manuscript. JF: data acquisition, data analysis and interpretation, drafting manuscript and final approval of manuscript.

**Funding** This research did not receive any specific grant from funding agencies in the public, commercial or not-for-profit sectors.

## Declarations

**Conflict of interest** The authors declare that they have no conflicts of interest.

**Ethical approval** Ethical approval for the study was provided by NHS Lothian Research and Ethics Committee.

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