

There are only minor changes in quality of life in patients with Scheuermann's disease

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ABSTRACT

INTRODUCTION: The impact of Scheuermann's disease (SD) on health-related quality of life (HRQoL) is unclear. The aim of this study was to study HRQoL based on The Medical Outcome Study Short Form-12 (SF-12) in adult life in a group of SD patients.

MATERIAL AND METHODS: A total of 46,418 twins were sent a questionnaire. 75.3% answered. Included in the questionnaire were questions from the SF-12. We compared SF-12 values in SD patients and control patients. We also identified 259 "non-concordant" twin pairs and compared SF-12 values from the SD twin with values from the healthy twin.

RESULTS: In all, 943 persons were identified to have SD, leaving 33,064 persons in the control group. SF-12 Physical Component Summary (PCS) (mean (standard deviation)) was found to be 50.50 (9.89) in SD and 53.21 (8.00) in controls ($p < 0.001$), and SF-12 Mental Component Summary (MCS) was found to be 51.52 (8.49) in SD and 51.81 (8.45) in controls ($p = 0.71$). In the non-concordant twin pairs, SF-12 PCS was found to be 50.74 (9.87) in SD and 52.74 (8.84) in controls ($p < 0.001$), and SF-12 MCS was found to be 53.91 (8.19) in SD and 53.72 (8.70) in controls ($p = 0.64$).

CONCLUSION: SF-12 PCS is moderately, though highly statistically significantly worse in SD patients than in controls in this big group of twins. We found no statistical difference in SF-12 MCS between the two groups. SD patients evaluate their physical health worse and their mental health like controls.

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In the literature, patients suffering from Scheuermann's disease (SD) have been reported to experience more back pain and other back-related constraints than the background population [1-4]. In addition, patients suffering from SD have been reported to experience more cosmetic problems [5] and a higher share than the background population experience psychological issues [6].

In SD as well as in many other diseases, the interest in assessing the patients' perceived health status has increased in recent years. Health-related quality of life

(HRQoL) assessments have become a significant outcome parameter in the medical literature; and the Medical Outcome Study Short Form-12 (SF-12) as well as the Medical Outcome Study Short Form-36 (SF-36) have become widely accepted as reliable tools. Both the SF-12 and the SF-36 have been cross-validated by Gandek et al for assessment of HRQoL in European countries including Denmark [7].

The impact of SD on HRQoL in patients remains unclear. We have been unable to find publications on HRQoL measured by the SF-12 or the SF-36 in SD.

The aim of this study was to explore the HRQoL based on the SF-12 in adult life in a group of patients with self-reported SD.

MATERIAL AND METHODS

The Danish Twin Registry is considered one of the most comprehensive registers of twins in the world holding more than 73,000 pairs born between 1870 and 2001 [8]. Zygosity of these twins was established based on four questions about similarity and mistaken identity with an accuracy of over 95% [9-11].

Twins are either mono-zygotic (MZ) or di-zygotic (DZ). MZ twins have identical segregating genes, while DZ twins, like siblings, share an average 50% of their genes. Both MZ and DZ twin pairs share a common environment for which reason only genetic factors account for a higher similarity in MZ than in DZ twin partners.

Twins born from 1931 through 1982 who were registered in the Danish Twin Registry with a permission for future contact were sent a questionnaire concerning diseases and health status in the spring of 2002 [12]. The questions included questions on SD.

A total of 46,418 twins received and 34,944 (75.3%) returned a questionnaire among whom 34,007 (97.3%) answered the question 'Have you been diagnosed with Scheuermann's disease by a doctor?' In Europe, including Denmark, Scheuermann's kyphosis is known under this name in the general population.

Included in the questionnaire were the standard SF-12 questions. Based on 12 questions, the SF-12 Physical Component Summary scale (SF-12 PCS) and the SF-12 Mental Component Summary scale (SF-12 MCS) are calculated using the SF Health Outcomes Scoring Software. The SF-12 was developed in 1994 by Ware et al as a sub-

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set of the SF-36 providing physical (PCS-12) and mental (MCS-12) summary health measures on two scales from 0-100 with a mean value of 50 in healthy persons [13]. The SF-12 PCS achieved a multiple R square of 0.911 in the prediction of the SF-36 PCS and, correspondingly, the SF-12 MCS achieved a value 0.918 in the prediction of the SF-36 MCS [13]. We compared the SF-12 values from SD patients to those of the control group.

We also identified “non-concordant” twin pairs (i.e. twin pairs in which one twin has SD and the other twin has not). In these non-concordant twin pairs, we compared the SF-12 values from the SD twin with the values from the “healthy” twin.

Statistics

SF Health Outcomes Scoring Software was used to calculate the SF-12 PCS and the SF-12 MCS. When comparing SD patients with controls, data were found to be normally distributed and Student’s t-test and the χ^2 -test were used to assess difference between cases and controls (Stata/SE 8.0 for windows, Stata Corporation, 4905 Lakeway drive, College Station, TX 77845, USA). In the smaller “non-concordant” twin groups, data were not found to be normally distributed and Wilcoxon’s signed-rank test was used to assess the difference between the SD twin and the healthy twin. p-values < 0.05 were considered significant.

Trial registration: The present study was approved by the Scientific Ethical Committee in Denmark (no. 20010202).

RESULTS

Of the 46,418 subjects available for the survey, 34,944 (75.3%) returned the questionnaire representing individuals from 23,204 twin pairs. The overall sex distribution among the responders was 54.5% (n = 19,037) females and 45.5% (n = 15,907) males. 34,007 of the responders replied to the question “Have you been diagnosed with Scheuermann’s disease”, and 943 of these answered “Yes” to this question.

This identified a group of self-reported SD patients of 943 persons, and a control group consisting of the remaining 33,064 patients. We identified 259 “non-concordant” twin pairs (i.e. twin pairs in which one twin has SD).

Table 1 shows the SF-12 PCS and the SF-12 MCS in all identified SD patients compared with the controls.

Table 2 shows the SF-12 PCS and the SF-12 MCS in “non-concordant” twin pairs, where values in SD twins are compared with the values of healthy twins.

We tested the PCS and MCS in the whole group of answers regardless of SD to control for differences in the background population based on gender. We found the SF-12 PCS to be 50.74 (9.88) in females and 52.89 (8.62) in males (p < 0.001) and the SF-12 MCS was determined to be 51.86 (8.18) in females and 51.67 (8.49) in males (p = 0.9).

The overall self-reported prevalence of SD was 2.8% (95% confidence interval (CI) 2.6-3.0). Gender stratification yielded a female prevalence of 2.1% (95% CI 1.9-2.3) and a male prevalence of 3.6% (95% CI 3.2-4.1) ($\chi^2 = 67.8$, p < 0.0001). The prevalence of SD was not significantly different between the groups of MZ and DZ twins [12].

DISCUSSION

In the complete cohort counting 943 patients with self-reported SD, we saw a moderately, but highly statistically significantly inferior SF-12 PCS compared with the control group (Table 1). We saw no statistical difference in the SF-12 MCS between the SD responders and the 33,064 controls (Table 1). Furthermore, when comparing the 259 “non-concordant” twin pairs, we reproduced the same result. Here, we again found a moderately, though highly statistically significantly inferior SF-12 PCS and no statistical difference in the SF-12 MCS between the SD twins and healthy twins (Table 2). This comparison of non-concordant twins is, of course, an adjustment for age. At the same time, it is an adjustment for different “environment factors”, since twin pairs share a common environment.

TABLE 1

SF-12 PCS and SF-12 MCS in all identified Scheuermann’s disease patients compared to controls.

	n	SF-12 PCS, mean (standard deviation)	SF-12 MCS, mean (standard deviation)
Scheuermann’s disease	943	50.50 (9.89)	51.52 (8.49)
Controls	33,064	53.21 (8.00)	51.81 (8.45)
p-value		< 0.001	0.71

MCS = Mental Component Summary; PCS = Physical Component Summary; SF = Short Form.

TABLE 2

SF-12 PCS and SF-12 MCS in “non-concordant” twin pairs. Scheuermann’s disease twins compared to healthy twins.

	n	SF-12 PCS, mean (standard deviation)	SF-12 MCS, mean (standard deviation)
Scheuermann’s disease twin	259	50.74 (9.87)	53.91 (8.19)
“Healthy” twin	259	52.74 (8.84)	53.72 (8.70)
p-value		< 0.001	0.64

MCS = Mental Component Summary; PCS = Physical Component Summary; SF = Short Form.

It should be noted that the SF-12 was developed as a subset of the SF-36 providing physical and mental summary health measures (PCS-12 and MCS-12) in two scales scoring participants from 0-100 with a mean of 50 in healthy persons [13]. Both in the SD group and in the controls, all of the values are close to the mean of 50 (Table 1 and Table 2).

We have been unable to find earlier studies on HRQoL in SD measured by the SF-12, the SF 36 or any other of the acknowledged health surveys. To measure the HRQoL in patients with spinal disorders, several both disorder-specific and generic questionnaires have been used. In recent years, the Scoliosis Research Society-22 Questionnaire (SRS-22) and the SF-36 have been the most widely used [14, 15]. The SF-12 used in this study was derived from the SF-36 instrument and has proved useful for group comparisons involving generic health concepts not specific to any age, disease or treatment group [16]. We therefore found the SF-12 relevant to use in this study.

Since we have made no correlations for gender, it seems reasonable to ask if the lower score in PCS might be explained by the fact that there are more males than females in the SD group. To adjust for this, we tested the PCS and MCS scores in the whole group of answers regardless of SD. We found the SF-12 PCS to be significantly lower in females than in males, but observed no statistical difference in the SF-12 MCS between females and males. This means that if gender were to explain the difference in PCS, we should have expected the SD group to have higher and not, as observed, lower PCS scores than the controls since there are more males than females in the SD group. It supports our findings that this is not the case.

Patients suffering from SD have been reported to experience more cosmetic problems [5] and to be facing a higher share of psychological issues [6] than the background population. This study does not support these reports, since we found no significant difference in MCS between SD and controls.

Since the prevalence of SD was not significantly different between the groups of MZ and DZ twins, being MZ or DZ twin does not affect the disease. We think that the results from this large cohort of twins are representative for the general population. Furthermore, we think that the large number of patients and controls in this cohort provides us with accurate figures. Had the cohort been smaller, we would not have found a statistically significant difference in SF-12 PCS.

The primary challenge in this study was the identification of the patients. Although this was done via a questionnaire, the case definition was not simply self-reported since the disorder had to be diagnosed by a doctor. It should therefore be kept in mind that the patients



Front page of the questionnaire.

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would have to have experienced symptoms or have had cosmetic complaints to seek a doctor in the first place. First, the patients would have to recall that they were diagnosed with SD; second, patients with unrecognised kyphosis may not have sought medical attention and the diagnosis would not have been made; third, some of these patients were born in the 1930s and 1940s and for those patients the diagnosis may not have been made since SD was first described in 1920. Obviously, there is therefore a risk that sub-clinical cases with no symptoms or cosmetic complaints are underestimated in this study.

Based on this study, we think that the appropriate information to the adolescent SD patient and his or her relatives is that the spine, though kyphotic, is as strong and healthy as any non-SD person's spine and fully capable of coping with the strains of a normal life. This information may be especially important in the mild and moderate curves, which are by far the most common, with a view to avoid stigmatising the patients and the relatives and induce unnecessary spinal morbidity.

CONCLUSION

We found no statistically significant difference in perceived mental health status; but in perceived physical health status, we found a significant difference between the two groups. Patients with self-reported SD scored moderately, but highly significantly worse on physical status than controls in this large cohort of Danish twins. Our interpretation of the present study is that SD does,

in fact, affect the HRQoL with reference to the patients' physical status. The influence of SD on the HRQoL is, however, very modest and in our opinion not clinically significant.

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