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Health-related quality of life in twins with clubfoot

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ABSTRACT

Introduction: Clubfoot is one of the most common congenital orthopedic conditions requiring treatment. The aim of this study was to examine the health-related quality of life (HRQoL) in twins with congenital clubfoot (CTEV). **Materials and Methods:** All 46,418 twins registered in the Danish Twin Registry born from 1931 to 1982 were sent a questionnaire. Included in the questionnaire were standardized questions for The Medical Outcome Study Short Form-12 (SF-12). From these questions, we calculated the SF-12 Physical Component Summary scale (SF-12 PCS) and the SF-12 Mental Component Summary scale (SF-12 MCS). These SF-12 values were compared to a control group and to the healthy twin in the twin pairs where only one twin has CTEV. **Results**: Of the 46,418 twins who received the questionnaire 34,944 (75%) returned it and 34,485 (99% of the responders) answered the question "Were you born with clubfoot?" The overall sex distribution among the responders was 55% (n = 19,037) females and 45% (n = 15,907) males. 94 reported to have clubfoot giving a self-reported prevalence of 0.27% (95% confidence interval 0.22-0.34%). There were 37 twin pairs where both twins answered all questions. There was no difference in the SF-12 PCS between genders (P = 0.37). There was a significant difference in SF-12 PCS though were the controls have a higher score than the CTEV twins, 53.10 versus 50.18 (P = 0.001). There was no difference in the SF-12 MCS (P = 0.202). However, the male CTEV twins had a higher score in SF-12 MCS than their female counterparts, 51.32 versus 48.47 (P = 0.047). There was no difference in SF-12 PCS or SF-12 MCS within the twin pairs (P = 0.32) and (P = 0.15). **Conclusion:** HRQoL from SF-12 PCS in self-reported CTEV was significantly worse than in controls in a big cohort of Danish twins. There was no difference in SF-12 MCS between the two groups. This indicates that patients with CTEV evaluate their physical health as worse than the control group. CTEV patients evaluate their mental health like the background population. However, female CTEV patients have a lower SF-12 MCS than their male counterparts. We found no difference in SF-12 PCS or SF-12 MCS between the CTEV patient and the healthy twin in the twin pairs where only one twin has CTEV.

KEY WORDS: Clubfoot, twins, HRQoL, health-related quality of life, SF-12

INTRODUCTION

Congenital clubfoot (CTEV) is evident both visually and functionally before and to some extends after treatment. This being more evident in patients treated surgically, before the current non-surgical techniques [1]. In CTEV as well as in many other diseases the interest in assessing the patients perceived health status has increased in the recent years. The so-called health-related quality of life (HRQoL) assessments have become a significant outcome parameter in the medical literature. The 12-item short form questionnaire SF-12® Health Survey (SF-12) has been widely accepted as a reliable tool for this purpose [2].

In a twin study, it is possible to compare a twin born with CTEV with a control group but also with the healthy twin, thus controlling for parental age, socioeconomic status at pregnancy and birth and with regard to monozygotic (MZ) twins for genes. This is a powerful design. The aim of this study was to examine HRQoL in twins with CTEV.

MATERIALS AND METHODS

The Odense based Danish Twin Registry contains data on all approximately 85.000 twin pairs born in Denmark over the last 140 years. It was the first population-based twin registry and is one of the largest existing today [3]. Zygosity of these twins is established based on four questions about similarity and mistaken identity with an accuracy of over 95% [3-7]. Twins born from 1931 through 1982 (between 20 and 71 years old) registered in the Danish Twin Registry and having consented to future contact were sent a questionnaire on diseases and health status in the spring of 2002. A total of 46,418 twins received and 34,944 (75%) returned a questionnaire of which 34,485 (99% of the responders) answered the question "Were you born with clubfoot?" The overall sex distribution among the responders was 55% (n = 19,037) females and 45% (n = 15,907) males.

Included in the questionnaire was the standardized questions for The Medical Outcome Study Short Form-12 (SF-12). The SF-12 is a questionnaire developed in 1994 as a subset of the SF-36 providing physical and mental summary health measures (PCS-12 and MCS-12) in two scales from 0 to 100 with a mean of 50 and a standard deviation of 10 [8]. SF-12 consists of 12 standard questions measuring health from the patient's point of view. SF-12 divides HRQoL in an SF-12 PCS and SF-12 MCS and is scored so that a high score indicates better function. In order to calculate SF-12 PCS and SF-12 MCS the answers to the 12 questions about HRQoL are scored in a complex algorithm.

We calculated the SF-12 PCS and the SF-12 MCS using the SF Health Outcomes Scoring Software.

SF-12 values were compared between CTEV patients and the rest of the cohort acting as controls and also with the healthy twin within a twin pair as the matched control.

The present study was approved by all Scientific-Ethical Committees of Denmark (case file 20010202).

Statistical Analysis

The prevalence of self-reported CTEV was calculated using the total number of answers as the general population.

Since the data were non-normally distributed but paired we used Wilcoxon matched-pairs signed-ranks test to assess differences between clubfoot- and healthy twins (Stata/SE 8.0 for windows, Stata Corporation, 4905 Lakeway drive, College Station, TX 77845, USA). $P < 0.05$ was considered significant.

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RESULTS

Totally, 46,418 twins received and 34,944 (75%) returned the questionnaire. A total of 34,485 twin individuals (99% of the responders) answered the question "Were you born with clubfoot?" 94 answered "yes" to the above question giving an overall self-reported the prevalence of congenital clubfoot of 0.0027 (95% confidence interval 0.0022-0.0034). A total of 29,596 (86%) twin individuals answered the SF-12. Of the clubfoot twins, 80 answered all SF-12 questions. There were 41 complete (both twins answered the question) twin pairs including four concordant pairs (both twins in a pair affected) two MZ and two dizygotic. These four twin pairs were excluded from the within the twin pair analysis. We had 37 complete twin pairs, where only one had CTEV in each pair for the SF-12 analysis.

SF-12 PCS and SF-12 MCS for all CTEV twins versus all healthy twins are presented in Table 1. There is a significant difference in PCS was the healthy twins has a higher score than the CTEV twins, 53.10 versus 50.18 (P = 0.001). There is no difference in the MCS (P = 0.202).

SF-12 PCS and SF-12 MCS for CTEV versus healthy twin (within a twin pair) are presented in Table 2. There is no difference in SF-12 PCS or SF-12 MCS within the twin-pairs (P = 0.32 and P = 0.15).

There was a gender difference in female CTEV patients has a lower SF-12 MCS than males 48.47 versus 51.32 (P = 0.047). There was no difference in the SF-12 PCS between genders (P = 0.37) [Table 3].

Table 1: SF-12 PCS and the SF-12 MCS compared in twins born with CTEV and healthy controls

All	*N*	Mean (SD)	
		SF-12 PCS	SF-12 MCS
Clubfoot	80	50.18 (11.19)	50.58 (10.52)
Controls	29,516	53.10 (8.11)	51.79 (8.47)
		P=0.001	*P*=0.209

SD: Standard deviation, MCS: Mental Component Summary scale, PCS: Physical Component Summary scale, CTEV: Congenital clubfoot

Table 2: SF-12 PCS and the SF-12 MCS in twins born with CTEV and their healthy twins

Twin pairs	*N*	Mean (SD)	
		SF-12 PCS	SF-12 MCS
Clubfoot twins	37	50.74 (10.50)	49.89 (10.98)
Healthy twins	37	54.41 (6.08)	51.71 (8.99)
		P=0.32	*P*=0.15

SD: Standard deviation, MCS: Mental Component Summary scale, PCS: Physical Component Summary scale, CTEV: Congenital clubfoot

DISCUSSION

Outcome measures in the treatment of CTEV are many and of various kinds. The aim of this study was to examine HRQoL in twins with congenital CTEV using the SF-12. In this cohort of 80 CTEV patients, we found a slight though statistically significant poorer SF-12 PCS when comparing to the 29,516 controls. We found no difference in SF-12 MCS.

In 2006 Dobbs *et al*. published a study on 45 patients with CTEV. Using SF-36 they found that CTEV patients at 30 years follow-up had a lower physical score but equal mental score as a published norm in the general United States population [9].

Wallander *et al*. published a similar study in 2009 on 83 CTEV patients. Male CTEV patients were found to have a higher score than female CTEV patients both in the physical and the mental score. There was no difference between the male CTEV patients and the norm group in the physical score but the female CTEV patients scored lower. There was no difference between the female CTEV patients and the norm group in the mental score but the male CTEV patients scored higher [10].

Both these studies have used The Medical Outcome Study Short Form-36 (SF-36) and not the SF-12. The SF-12 used in this study was derived from the SF-36 instrument that has proved useful for group comparisons involving generic health concepts not specific to any age, disease or treatment group [11]. Even though SF-12 and SF-36 are not the same, SF-12 PCS achieved a multiple R^2 = 0.911 in the prediction of the SF-36 PCS and SF-12 MCS in the same way 0.918 in the prediction of the SF-36 MCS [8].

Our interpretation of the present study is that CTEV in this cohort, in fact, does affect the HRQoL with reference to the physical status of the patients. The clubfeet in this cohort were treated many years ago with the surgical practices of that time, with extensive posteromedial releases. Several studies have indicated that surgical releases led to long-term complications [9,12]. None of the clubfeet in this study was treated with the current non-surgical techniques, which might influence the SF-12 PCS differently.

The difference in SF-12 PCS doesn't influence the SF-12 MCS in these patients since we found no difference in SF-12 MCS. This positive finding suggests that the CTEV patients adapt to the lower physical ability and learn to cope with the complications of the clubfoot.

Table 3: SF-12 PCS and the SF-12 MCS in male and female twins born with CTEV

Twin pairs	*N*	Mean (SD)	
		SF-12 PCS	SF-12 MCS
Male	37	52.39 (9.32)	51.32 (12.83)
Female	37	49.10 (11.57)	48.47 (8.87)
		P=0.37	*P*=0.047

SD: Standard deviation, MCS: Mental Component Summary scale, PCS: Physical Component Summary scale, CTEV: Congenital clubfoot

When comparing the 37 complete (both twins answering all questions) non-concordant twin pairs, we found no statistical difference in SF-12 PCS or SF-12 MCS. The means are different with the SF-12 PSC in the CTEV twins shoving a lower tendency, but this is not statistically significant.

There is a gender difference since the female CTEV patients had a lower SF-12 MCS than the males. This is consistent with the finding in previous studies [10]. One explanation could be cosmetic problems with the smaller calf and foot and scaring from the operations, being a bigger problem for the female clubfoot patients.

This study has several limitations. The main challenge in questionnaire-based studies is the identification of the patients. In the present study on CTEV, we consider this to be a reliable method since this distinct disorder is not easily overlooked or mistaken for other frequent disorders. We do not expect patients to be unaware of having had CTEV however well corrected. <50% of CTEV will be corrected in one operation, around 80% in two operations and approximately 90% after three operations [12]. Long-term follow-up after surgical correction, indicates that many patients suffer from pain, limping, and overall reduced foot function [9].

Another limitation is the lack of knowledge as to initial CTEV severity and treatment. Within the CTEV spectrum, there are different severities and certainly so with different treatments. Even within a small country, being born with, and treated for, clubfoot is, therefore, many things and patients are probably not a homogenous group.

SF-12 has been widely accepted as a reliable tool to quantify HRQoL. However, one could speculate if the outcomes of the questionnaire are sensitive enough when comparing patients with congenital clubfoot and a background population and to the health twin in twin pairs. In both the clubfoot twins and the healthy twins all of the values are relatively close to the mean of 50. Finally, we don't know the clinical implications of the differences in SF-12 MCS and SF-12 PCS.

CONCLUSION

Since CTEV patients treated surgically in many cases, experience reduced foot function and other problems it is not surprising that patients with self-reported CTEV score significantly worse in SF-12 PCS than controls in this big cohort of Danish twins. However, this physical problem doesn't seem to influence the mental wellbeing of the CTEV patients.

We found no difference in SF-12 MCS between CTEV patients and the controls. Maybe this is due to the fact that in congenital disease the patients to some degree "learn to live with it" and/or to some extend abstain from various physical activities. Female clubfoot patients had a lower SF-12 MCS than male patients.

In this cohort of 80 CTEV patients, we found a slight though statistically significant, poorer SF-12 PCS when comparing to

the 29,516 controls. We found no difference in SF-12 MCS.

When comparing the 37 complete (both twins answering all questions) non-concordant twin pairs, we found no statistical difference in SF-12 PCS or SF-12 MCS. The means are different with the SF-12 PSC in the CTEV twins shoving a lower tendency, but this is not statistically significant. In both the clubfoot twins and the healthy twins all of the values are relatively close to the mean of 50.

There was a gender difference in female CTEV patients has a lower SF-12 MCS than males 48.47 versus 51.32. There was no difference in the SF-12 PCS between genders.

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