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Improving Quality of Life With Dynamic Compression Bracing in Patients With Pectus Carinatum



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ABSTRACT

Background: Patients with pectus carinatum have lower quality of life and self-esteem than their peers. We assessed the impact of dynamic compression system bracing on quality of life in patients with pectus carinatum.

Methods: We conducted a prospective cohort study on patients aged 10–21 years. We assessed quality of life using the Child Health Questionnaire-87, the State-Trait Anxiety Inventory-6, the World Health Organization Quality of Life-BREF, the 36-Item Short Form Survey, and the Single-Step Questionnaire adapted for pectus carinatum.

Results: Between March 2013 and March 2016, 225 patients treated with dynamic compression system bracing were included. Patients showed improvements across the overall scores of the 36-Item Short Form Survey ($\Delta 7.7$ (2.9–12.4)), Single-Step Questionnaire ($\Delta 4.1$ (2.0–6.3)) and three out of four World Health Organization Quality of Life-BREF domains (physical health ($\Delta 8.7$ (3.7–13.7)), psychological health ($\Delta 11.8$ (6.1–17.5)), environment ($\Delta 5.7$ (0.2–11.3))). No changes across the Child Health Questionnaire-87 overall score were observed ($\Delta 5.5$ (–0.5–11.5)). Most improvement occurred within six to twelve months after treatment initiation, stabilizing thereafter. Anxiety scores on the State-Trait Anxiety Inventory-6 did not improve ($\Delta 0.5$ (–0.1–1.2)). Scores on physical complaints, pain, psychological health and self-esteem/self-image improved across all questionnaires. In contrast to the successfully treated group, the unsuccessfully treated group showed no improvement on any of the questionnaires. Most patients (87.2 %) would choose bracing again, 94.9 % of patients were satisfied with the treatment.

Conclusions: Dynamic compression system bracing improves quality of life, reduces physical complaints and pain and boosts psychological health and self-esteem in patients with pectus carinatum.

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1. Introduction

Pectus carinatum (PC) is a pediatric condition marked by abnormal overgrowth of costal cartilage which results in protrusion of the sternum. Incidence varies from 0.04 to 0.7 % in the current literature [1,2]. Almost 50 % of the patients feel embarrassed about their chest and experience occasional bullying. This may lead them

to avoid activities in which their chest is visible such as swimming. It may cause psychological distress, a disturbed body image and an overall reduced quality of life (QoL) [2–4]. Additionally, patients often report various physical complaints, primarily respiratory and cardiovascular, but also pain [5,6]. Currently, dynamic compression system bracing (DCS-bracing) is the treatment option of first choice, with good results [2]. Despite these results, there is limited

Abbreviations: 95%CI, 95 % Confidence Interval; CHQ-87, Child Health Questionnaire-87; DCS-bracing, Dynamic Compression System Bracing; IQR, Interquartile Range; MCS, Mental Component Score; METC, Medical Ethics Review Committee; PCS, Physical Component Score; PC, Pectus Carinatum; QoL, Quality of Life; SF-36, Short Form Survey 36; SSQ, Single Step Questionnaire; STAI-6, Six-item State-Trait Anxiety Inventory; WHOQOL-BREF, World Health Organization Quality of Life Brief Version.

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knowledge about the effect of DCS-bracing on improvements in QoL, self-esteem and psychological distress. Only three studies have addressed (facets of) QoL after treatment; however, they used different braces and bracing protocols [7–9]. Two studies report on improvement of QoL after surgical correction of PC [10,11]. The lack of studies on this topic is particularly surprising as a reduced quality of life is the primary reason for seeking treatment.

Therefore, we evaluate the changes in QoL in patients with PC treated using DCS-bracing, with emphasis on physical complaints, pain, psychological health and self-esteem/self-image.

2. Materials and methods

2.1. Patients and study design

We conducted a prospectively cohort study of patients who commenced DCS-bracing therapy between March 2013 and March 2016 at the Amsterdam Pectus Centre. Patients aged ten or older

with sufficient knowledge of the Dutch language were included. A Medical Ethics Review Committee (METC) official waiver of ethical approval was granted by the METC of the Amsterdam Medical Center. Informed consent was obtained from all patients (or their parents) included in this study.

2.2. Treatment protocol

At the Amsterdam Pectus Centre, patients with pectus carinatum (PC) are treated using the FMF Dynamic Compression System brace (Pampamed). Pediatric surgeons handle both intake and follow-up appointments, following the treatment protocol outlined in Fig. 1. Patients are advised to delay treatment until after the onset of their growth spurt. The pressure of initial correction is assessed using a pressure device that measures the average force required to correct the chest deformity. Patients are advised to wear the brace as much as possible, and we encourage them to wear the brace at night to extend their treatment hours more comfortably.

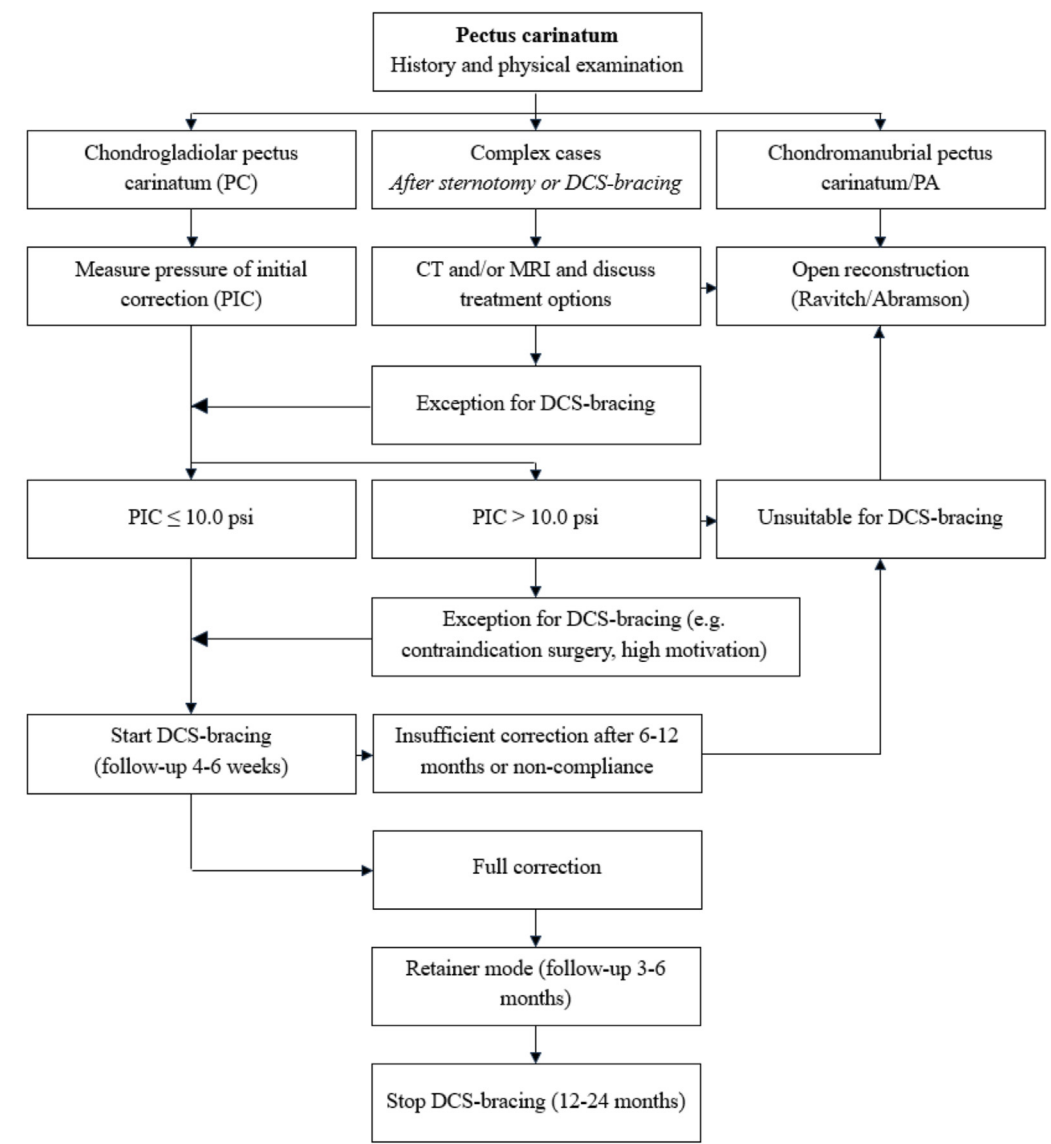


Fig. 1. Treatment protocol (DCS-bracing = Dynamic Compression System bracing, PA = Pectus Arcuatum, PC = Pectus Carinatum, PIC = Pressure of Initial Correction).

Treatment is deemed successful when a satisfactory outcome is achieved, as determined in consultation with the clinician.

2.3. Questionnaires

Patients were divided into three age groups (10–16y, 16–18y and >18y), to meet the validation criteria for the various questionnaires, with distinct questionnaires administered to each age group. Patients younger than ten years were excluded. Questionnaires were administered at the start of treatment and at one, six, twelve, eighteen and 24 months thereafter.

The Single Step Questionnaire (SSQ) (A), originally developed to assess QoL in patients with PE after the Nuss procedure, was used to assess change in QoL and satisfaction during treatment [12]. The questionnaire was adapted in our center to fit PC patients and was used with all subjects. All items, except for three, were scored on a 5-point Likert scale.

Anxiety was assessed using the Six-item State-Trait Anxiety Inventory (STAI-6) (A) [13]. Items were scored on a 4-point Likert scale. The total score was displayed on a 0–24 scale.

The evaluation of QoL in the younger patients, aged ten to eighteen years, was done with the Child Health Questionnaire-87 (CHQ-87) (A). The CHQ-87 encompasses different domains, covering physical, emotional, and social well-being domains, including specific inquiries regarding self-esteem and self-image perception [14]. Items were scored using a 4- or 5-point Likert scale. Domain scores were converted to a standardized 0–100 scale.

The World Health Organization Quality of Life Brief Version (WHOQOL-BREF) (A), a condensed version of the World Health Organization Quality of Life-100 (WHOQOL-100), was used to assess QoL in patients aged sixteen years and older across four key domains, being physical health, psychological health, social relationships and environment. Additionally, it assesses two overarching aspects (QoL and health satisfaction in general). To enhance our study's scope, we added the facet body image and facet pain of the WHOQOL-100. Items were scored on a 5-point Likert scale. Domain scores were converted to a standardized 0–100 scale [15].

Besides the WHOQOL-BREF the Short Form Survey 36 (SF-36) (A), a 36-item questionnaire covering eight domains of health, was used for patients aged sixteen years and older. This questionnaire also focusses on the limitations in daily life due to physical health or emotional problems [16]. We calculated the physical and mental component scores (PCS and MCS) besides the total score of the SF-36 to measure QoL, as previously advised [17,18]. Items were scored on a combination of Likert scales and binary scales. Domain scores were converted to a standardized 0–100 scale.

Scores of all items in the questionnaires were converted to positive scores to standardize the scoring. Higher scores indicated a better QoL.

2.4. Statistical analysis

Data were analyzed using IBM SPSS Statistics 28.0. Descriptive measurements were utilized to characterize the study population. Changes in QoL primary and secondary outcomes were analyzed using linear mixed models, given that this method is ideal for analyzing repeated measures with missing data [19]. The linear mixed models only included time, which was treated as a categorical outcome variable represented by five dummy variables.

We used 95 % Confidence Intervals (95%CI) to determine statistical significance. A 95 % Confidence Interval that does not include the null value indicates that the result is statistically significant at the $P < 0.05$ level. The change in questionnaire scores is reported in the text as the absolute difference with 95 % Confidence

Intervals, formatted as (absolute difference (95%CI)), e.g., ($\Delta 5.5$ (-0.5 – 11.5)).

2.5. Outcomes

Primary outcomes were changes in QoL due to DCS-bracing. Secondary outcomes were changes in anxiety scores, changes in subdomains of the questionnaires, especially physical complains, pain, psychological health and self-esteem/self-image, and differences in outcomes between successfully and unsuccessfully treated patients.

3. Results

Of all patients ($N = 260$) treated in the study period 225 patients were included. Thirty-five patients were excluded either because of insufficient knowledge of the Dutch language, or they did not return any of the questionnaires. Baseline characteristics are listed in Table 1. Of all patients, 92.0 % were male ($n = 207/225$), median age was 15.0 (interquartile range (IQR) 13.0–16.0) years and median treatment time was 16.0 (IQR 12.0–23.0) months. All patients were diagnosed with PC, and none with pectus arcuatum (also known as Type 2 PC or Currarino-Silverman syndrome). None of the patients were still actively treated at the time of evaluation, 78.2 % ($n = 176/225$) finished treatment successfully, 13.3 % ($n = 30/225$) finished treatment unsuccessfully and 8.4 % ($n = 19/225$) were lost to follow-up. The number of respondents for each questionnaire at each time point is detailed in Table 2. Initial response rates ranged from 62.7 % to 77.3 % at the zero-month mark, with a gradual decline over time.

3.1. CHQ-87

Table 3 shows the absolute and relative changes in overall scores of all questionnaires, while Fig. 2 visually represents these changes in a graph to provide a clear overview. Total scores on the CHQ-87 did not change over the two years follow-up ($\Delta 5.5$ (-0.5 – 11.5)), although it increased from 76.1 to 81.6 points. More than half of this improvement occurred within the first six months of treatment. There were changes in the following subdomains: physical functioning ($\Delta 1.9$ (0.3–3.6)), role/social limitations due to behaviour ($\Delta 2.2$ (0.6–3.8)), pain ($\Delta 6.3$ (1.1–11.6)), mental health ($\Delta 3.0$ (0.1–5.9)), and self-esteem ($\Delta 3.4$ (0.2–6.4)). There were no changes in the subdomains general health ($\Delta 3.9$ (-1.0 – 8.8)), role/social limitations due to physical health ($\Delta 1.9$ (-0.0 – 3.8)), role/social limitations due to emotional health ($\Delta 2.3$ (-0.4 – 5.0)), behaviour ($\Delta 2.4$ (-0.1 – 4.9)), family cohesion ($\Delta 16.9$ (-1.5 – 35.2)),

Table 1
Baseline characteristics.

	All patients (N = 225)
Sex ^b	
Male	207 (92.0)
Female	18 (8.0)
Age (years) ^a	15.0 (13.0–16.0)
<16 years ^b	156 (69.3)
16–18 years ^b	57 (25.3)
>18 years ^b	2 (0.9)
Treatment time (months) ^a	16.0 (12.0–23.0)
Results ^b	
Treatment successful	176 (78.2)
Treatment not successful	30 (13.3)
Lost to follow-up	19 (8.4)

^a Nonparametric variables; expressed as median (IQR).

^b Data displayed as n (%).

Table 2
Responses per questionnaire.

	0m ^a	1m ^a	6m ^a	12m ^a	18m ^a	24m ^a
CHQ-87 (N = 223 ^b)	161	123	73	69	30	4
SF-36 (N = 59 ^b)	38	26	20	6	8	12
WHOQOL-BREF (N = 59 ^b)	37	31	23	15	8	15
STAI-6 (N = 225 ^b)	174	127	85	76	43	52
SSQ (N = 225 ^b)		127	97	68	43	75

CHQ-87 = Child Health Questionnaire-87, m = months, N/n = number, SF-36 = Short Form Survey 36, SSQ = Single Step Questionnaire, STAI-6 = Six-item State-Trait Anxiety Inventory, WHOQOL-BREF = World Health Organization Quality of Life Brief Version.

^a n of responses.

^b Number of patients eligible for this questionnaire at 0 months.

or family activities ($\Delta 0.9$ (-2.8 – 4.5)). The positive change in health increased over the two years from 3.2 to 7.1 points ($\Delta 4.0$ (2.3 – 5.6)).

3.2. SF-36

Total scores on the SF-36 did change over two years ($\Delta 7.7$ (2.9 – 12.4)), increasing from 83.4 to 91.0 points. QoL improved until six months after treatment initiation and then remained stable without decline. The PCS ($\Delta 0.5$ (-4.4 – 3.4)) and the MCS ($\Delta 1.3$ (-5.6 – 3.0)) did not change over the treatment period.

There were changes in the subdomains physical functioning ($\Delta 4.4$ (1.2 – 7.6)), role of limitations due to physical health ($\Delta 11.2$ (3.1 – 19.4)), energy/fatigue ($\Delta 10.5$ (4.2 – 16.8)), emotional well-being ($\Delta 7.4$ (2.2 – 12.7)), social functioning ($\Delta 11.9$ (5.9 – 17.9)), pain ($\Delta 9.0$ (3.5 – 14.4)), and general health ($\Delta 10.5$ (3.1 – 18.0)). There were no changes in the subdomain role of limitations due to emotional problems ($\Delta 8.8$ (-1.0 – 16.5)). The positive change in health was stable during the two years and did not change ($\Delta 4.2$ (-3.1 – 11.5)).

Table 3
Absolute and relative change in (overall) scores of questionnaires.

	0m	1m	6m	12m	18m	24m
CHQ-87						
Mean ^a	76.1 (6.8)	76.6 (6.9)	79.3 (4.4)	79.6 (4.2)	79.1 (4.3)	81.6 (7.1)
Difference from baseline ^b		0.5 (0.7)	3.2 (4.2)	3.5 (4.6)	3.0 (3.9)	5.5 (7.2)
SF-36						
Mean ^a	83.4 (13.7)	85.6 (13.2)	91.3 (7.5)	92.7 (5.3)	89.8 (8.0)	91.0 (10.5)
Difference from baseline ^b		2.2 (2.6)	7.9 (9.5)	9.3 (11.2)	6.4 (7.7)	7.7 (9.1) ^c
WHOQOL-BREF						
Physical health						
Mean ^a	78.4 (12.1)	79.4 (15.4)	87.1 (10.8)	87.4 (10.8)	86.8 (10.4)	87.1 (10.1)
Difference from baseline ^b		1.0 (1.3)	8.7 (11.1)	9.0 (11.5)	8.4 (10.7)	8.7 (11.1) ^c
Psychological health						
Mean ^a	71.7 (13.5)	77.1 (14.8)	82.5 (11.7)	83.3 (9.5)	81.9 (9.2)	83.5 (9.3)
Difference from baseline ^b		5.4 (7.5)	10.8 (15.1)	11.6 (16.2)	10.2 (14.2)	11.8 (16.5) ^c
Social relationships						
Mean ^a	79.8 (14.1)	78.2 (15.5)	82.2 (11.7)	81.4 (14.2)	83.9 (11.9)	81.0 (12.0)
Difference from baseline ^b		-1.6 (-2.0)	2.4 (3.0)	1.6 (2.0)	4.1 (5.1)	1.2 (1.5)
Environment						
Mean ^a	81.1 (10.3)	81.8 (13.0)	88.8 (10.6)	88.4 (8.9)	86.9 (11.6)	86.9 (9.3)
Difference from baseline ^b		0.7 (0.9)	7.7 (9.5)	7.3 (9.0)	5.7 (7.1)	5.7 (7.1) ^c
STAI-6						
Mean ^a	21.0 (2.7)	21.3 (2.6)	21.9 (1.7)	21.8 (2.2)	21.2 (2.3)	21.5 (2.4)
Difference from baseline ^b		0.3 (1.4)	1.0 (4.3)	0.8 (3.8)	0.3 (1.0)	0.5 (2.4)
SSQ						
Mean ^a		68.6 (7.3)	70.3 (7.3)	73.2 (7.4)	72.6 (7.9)	72.7 (4.1)
Difference from baseline ^b			1.7 (2.5)	4.6 (6.7)	4.0 (5.8)	4.1 (6.0) ^c

CHQ-87 = Child Health Questionnaire-87, m = months, SF-36 = Short Form Survey 36, SSQ = Single Step Questionnaire, STAI-6 = Six-item State-Trait Anxiety Inventory, WHOQOL-BREF = World Health Organization Quality of Life Brief Version.

^a Mean (SD).

^b Absolute difference from baseline (percentage change relative to baseline).

^c Statistically significant values (95 % Confidence Intervals calculated only for the 24-month data).

3.3. WHOQOL-BREF

On all domains most improvement was made in the first six months after treatment initiation, after that it remained stable. There were changes in the subdomains physical health ($\Delta 8.7$ (3.7 – 13.7)), psychological health ($\Delta 11.8$ (6.1 – 17.5)), environment ($\Delta 5.7$ (0.2 – 11.3)), pain ($\Delta 13.1$ (3.2 – 23.0)), body image ($\Delta 37.7$ (29.5 – 45.8)). General QoL ($\Delta 10.9$ (5.5 – 16.3)) and health satisfaction ($\Delta 10.7$ (4.3 – 17.1)) did also improve. There was no change in the subdomain social relationships ($\Delta 1.2$ (-4.5 – 6.9)).

3.4. STAI-6

Total scores on the STAI-6 did not change over two years ($\Delta 0.5$ (-0.1 – 1.2)), although it increased from 21.0 to 21.5 points over two years. Most improvement was made in the first six months, with a slight decrease in total scores after six months.

3.5. SSQ

Total scores on the SSQ did change over two years ($\Delta 4.1$ (2.0 – 6.3)), increasing from 68.6 to 72.7 points. QoL improved until twelve months after treatment initiation, after that it remained stable, without any significant decline. During treatment scores on exercise capacity ($\Delta 0.5$ (0.2 – 0.7)), satisfaction with body ($\Delta 0.3$ (0.1 – 0.5)), impact of DCS-bracing on social life ($\Delta 0.4$ (0.2 – 0.6)), limitations on social activity during bracing ($\Delta 0.5$ (0.1 – 0.8)), self-image after start of DCS-bracing ($\Delta 0.5$ (0.2 – 0.8)), pain in general ($\Delta 0.1$ (0.0 – 0.4)), and improvement of the thorax ($\Delta 0.2$ (0.0 – 0.4)) increased.

Scores on general health ($\Delta 0.2$ (-0.0 – 0.4)), limitations on social activity before DCS-bracing ($\Delta 0.1$ (-0.2 – 0.4)), skin problems during treatment ($\Delta 0.1$ (-0.3 – 0.4)), self-image before start DCS-bracing ($\Delta 0.2$ (-0.3 – 0.7)), pain during the application of the DCS-

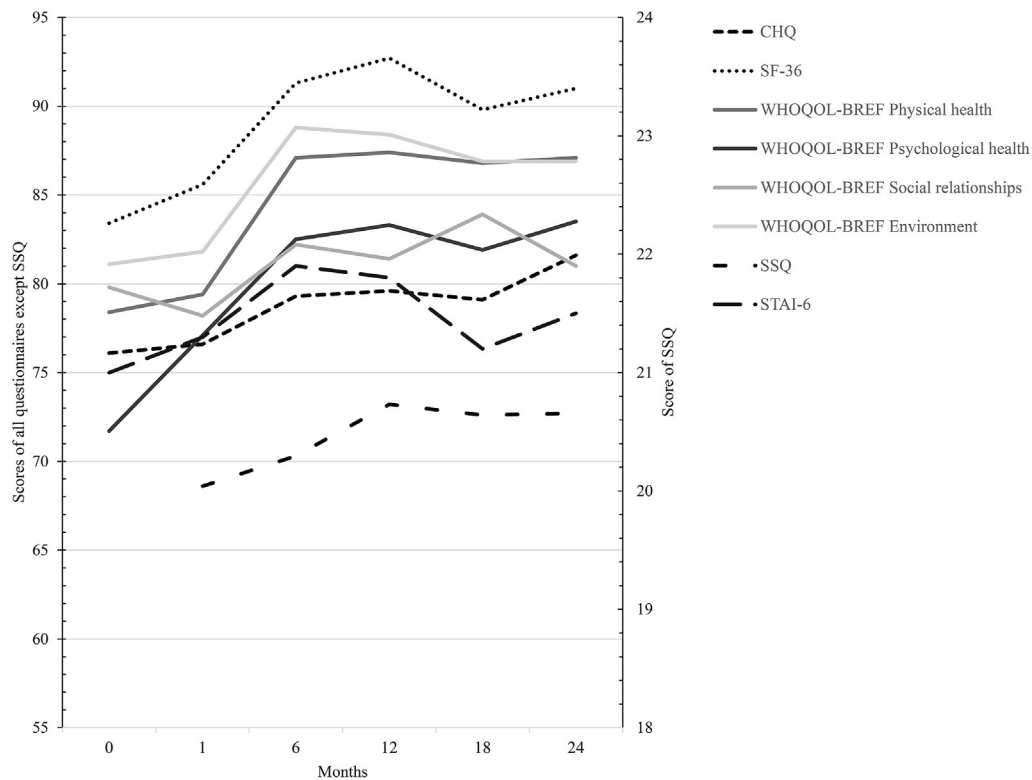


Fig. 2. Change in (overall) scores of questionnaires (CHQ-87 = Child Health Questionnaire-87, SF-36 = Short Form Survey 36, SSQ = Single Step Questionnaire, STAI-6 = Six-item State-Trait Anxiety Inventory, WHOQOL-BREF = World Health Organization Quality of Life Brief Version).

brace ($\Delta 0.1$ (-0.2 – 0.4)), pain during daily activities ($\Delta 0.1$ (-0.1 – 0.4)), satisfaction with the DCS-brace ($\Delta 0.2$ (-0.0 – 0.4)), and awareness of the DCS-brace during wearing ($\Delta 0.3$ (-0.0 – 0.5)) did not change.

Of all patients, 96.5 % ($n = 192/199$) was either satisfied (42.7 %, $n = 85/199$), very satisfied (34.7 %, $n = 69/199$) or extremely satisfied (19.1 %, $n = 38/199$) with DCS-bracing at the conclusion of their treatment or at their last assessment before dropping out. Additionally, 85.0 % ($n = 170/200$) of patients would opt for DCS-bracing again if they had to make the decision anew, 12.5 % ($n = 25/200$) had doubts and 2.5 % ($n = 5/200$) would definitely not choose DCS-bracing again.

3.6. Improvement in QoL and success of treatment

Overall scores on the SF-36 ($\Delta 9.0$ (3.6–14.5)), scores on the subdomains physical health ($\Delta 9.4$ (3.7–15.0)), psychological health ($\Delta 11.9$ (5.5–18.3)) and environment ($\Delta 6.4$ (0.3–12.6)) of the WHOQOL-BREF, and overall scores on the SSQ ($\Delta 4.6$ (2.4–6.7)) did change in the successfully treated group. Scores on the subdomain social relationships ($\Delta -0.5$ (-6.7 – 5.7)) and scores on the CHQ-87 ($\Delta 5.0$ (-0.6 – 10.6)) and the STAI-6 ($\Delta -0.5$ (-0.2 – 1.2)) did not change. In contrast, the unsuccessfully treated group showed no improvement in the total scores of any of the questionnaires (CHQ-87 ($\Delta 5.9$ (-2.1 – 13.9)), SF-36 ($\Delta -1.5$ (-15.9 – 12.9)), subdomains physical health ($\Delta -0.5$ (-17.2 – 16.2)), psychological health ($\Delta 6.4$ (-14.3 – 27.2)), social relationships ($\Delta -2.9$ (-27.7 – 22.0)) and environment ($\Delta -4.0$ (-26.4 – 18.3)) of the WHOQOL-BREF, STAI-6 ($\Delta -0.3$ (-2.3 – 1.7)) and SSQ ($\Delta -4.7$ (-11.9 – 2.5))). This led to overall lower scores in QoL in the unsuccessfully treated group compared to the

successfully treated group. Still, 65.8 % ($n = 25/38$) of the unsuccessfully treated group would opt for DCS-bracing again if they had to make the decision anew and 86.5 % ($n = 32/37$) was either satisfied or very satisfied with the treatment at their last assessment.

4. Discussion

The overall QoL of patients treated with DCS-bracing for PC increased across the SF-36 and the WHOQOL-BREF, but not on the CHQ-87. The largest change in QoL took place in the first six to twelve months after treatment initiation, after this period QoL stabilized. There was no improvement in anxiety scores across the STAI-6. Scores on physical complains, pain, psychological health and self-esteem/self-image improved across all questionnaires. Most patients (87.2 %) would choose DCS-bracing again and 94.9 % were satisfied with the treatment.

In contrast to the successfully treated group, the unsuccessfully treated group showed no improvement on any of the questionnaires.

4.1. Change in QoL

The QoL measured by the CHQ-87, did not change. This may be due to the age difference between patients completing the CHQ-87 (aged 14.0 (13.0–15.0)) and those completing the SF-36 and WHOQOL-BREF (aged 17.0 (16.0–17.0)). Concerns about appearance typically begin around puberty, which tends to start later in boys. In our population, 92 % of the patients are male. In a significant part of the younger group, it is likely that self-esteem concerns have not

yet developed, and it is mainly the parents who are worried about the condition rather than the children themselves, often leading parents to initiate treatment decisions. In contrast, the other group consists of patients who are clearly going through puberty, resulting in increased self-awareness and self-esteem issues, prompting them to seek treatment independently. Consequently, the younger group may have a higher baseline QoL since their QoL is probably not affected, which could explain the lack of significant change in QoL observed in this group. However, assessing this difference in baseline QoL is challenging due to the use of different questionnaires across the groups.

This is one of the first studies to report on changes in QoL, making it difficult to compare our results to existing literature. The only study examining overall QoL in patients with PC reported no significant change in overall health over a follow-up period of 41 weeks [8]. Nevertheless, literature is available on related topics such as anxiety, self-esteem, and treatment satisfaction.

4.2. Anxiety, physical complaints, pain, psychological health and self-esteem/self-image

Earlier research suggested that PC affects patients the most on their psychological health, self-image and self-esteem but also causes physical complaints and pain [3–6]. Every questionnaire showed improvement in the physical domain, with treatment leading to improvement of physical complaints, increased exercise capacity and energy. The same applies for pain, psychological health and self-esteem/self-image. There was no improvement on state anxiety scores.

These results are consistent with the scarcely available literature. Colozza et al. reports an improvement of both chest appearance and self-esteem (+17.8 %) in a small group of patients (n = 20) that were successfully treated with an external brace [7]. Fraser et al. report changes on anxiety/depression (–4 on a 10-point Likert scale) and appearance/body confidence (+6 on a 10-point Likert scale) in a study with 159 patients [8]. None of the studies reported on improvement of physical complaints or pain.

4.3. Treatment evaluation

In general, patients were very satisfied with the DCS-brace, 94.9 % of the patients was satisfied and 87.2 % of patients were confident that they would use the brace again. This is similar to another study, which reported a satisfaction rate of 94.8 % [9]. In another study the SSQ was also used, with a reported mean satisfaction of 3.7 (on a 5-point Likert-scale), comparable to our mean satisfaction of 3.8 after two years. They also reported that most patients would use the brace again [7]. Other studies used a single 5- or 10-point Likert scale to assess patients satisfaction, reporting 4.7 out of 5 [20] and 7–8 out of 10 [8,21].

Even when treatment fails, 65.8 % of patients would choose DCS-bracing as treatment again and 86.5 % of these patients were satisfied with the treatment. This highlights the overall positive perception of DCS-bracing as a treatment for PC.

4.4. Treatment duration and results

In one of our earlier studies in the same patient population, we demonstrated improvement in self-assessment of the chest wall, with scores increasing from 4.0 to 7.8 on a 10-point Likert scale within the first six months [6]. As seen in our QoL results, improvements stabilize after six months, suggesting that most progress occurs during this initial period, which patients can notably observe. This early improvement boosts their confidence, thereby enhancing QoL scores. Although the results on improvement of QoL

suggest that after six months not much progress is made, the remaining months of treatment, with a median duration of sixteen months, are crucial for maintaining the chest wall's position to prevent relapse.

4.5. Limitations

A limitation of this study is the gradual decline in response rates over time, which is anticipated as some patients complete treatment early and are subsequently lost to follow-up. Despite this, such attrition may still influence the robustness of the reported outcomes. Although the use of different questionnaires was necessary for our different aims (CHQ-87 for younger patients, SF-36 and WHOQOL-BREF for older patients, STAI-6 for anxiety, and SSQ for treatment satisfaction), questionnaire fatigue may have contributed to the decline in responses.

Additionally, the lack of a control group for comparison limits our ability to assess whether the post-treatment QoL is comparable to that of healthy individuals. Incorporating a control group in future studies would provide a stronger basis for evaluating the true impact of treatment on QoL in these patients.

Another limitation relates to not using Minimal Clinically Important Differences (MCID). These values for the WHOQOL-BREF and SF-36 have been established for chronic and severe illnesses, such as cancer, but not for pectus deformities [22]. Although some studies have cautiously applied these MCID values to less serious conditions, they typically involved populations of similar age to those in the original MCID studies and conditions more serious than pectus deformities [23]. In contrast, our study includes a significantly younger population with a much less severe condition, making the cautious application of these MCID values unsuitable in our opinion. Determining MCID values specifically for pectus deformities is necessary to accurately define what percentage increase in quality of life is clinically meaningful for this population.

5. Conclusions

DCS-bracing is a successful treatment in patients with PC, improving QoL in general and on different subdomains. DCS-bracing decreases physical complaints and pain and improves psychological health and self-esteem/self-image. The largest changes in QoL take place in the first six to twelve months after treatment initiation. Overall most patients are confident that they would choose DCS-bracing again and nearly all patients are satisfied with the treatment.

Previous communication

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Conflicts of interest

Sjoerd A. de Beer, contracted by the company Atricure, works as a proctor for cryotherapy in pectus patients. The other authors have no relevant financial or non-financial interests to disclose.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.jpedsurg.2024.161975>.

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