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# Pectus excavatum and pectus carinatum patients suffer from lower quality of life and impaired body image: a control group comparison of psychological characteristics prior to surgical correction<sup>☆</sup>

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## Abstract

Objective: The aim of this study was to evaluate the effects of anterior chest-wall deformities on disease-specific and health-related quality of life, body image, and psychiatric comorbidity prior to surgical correction. Methods: A total of 90 patients (71 with pectus excavatum, 19 with pectus carinatum) presenting themselves for pectus repair and 82 control subjects were recruited for this study. The objective severity of the deformity was determined through the funnel-chest index by Hümmer and the Haller index. Disease-specific quality of life was measured with the Nuss Questionnaire modified for Adults (NO-mA) and health-related guality of life was determined by the Short-Form-36 Health Survey (SF-36). Body image was assessed via the Body Image Questionnaire (FKB-20), the Dysmorphic Concern Questionnaire (DCQ), and a self-evaluation of the subjective impairment of the appearance. The Diagnostic Interview for Mental Disorders - Short Version (Mini-DIPS), the General Depression Scale (Allgemeine Depressionsskala, ADS), and a self-rating of self-esteem were used to evaluate general psychological impairment. Results: Compared with control group results, physical quality of life was reduced in patients with pectus excavatum, while mental quality of life was decreased in patients with pectus carinatum (p < 0.05). Body image was highly disturbed in all the patients and differed significantly from the control group (p < 0.01). Patients with pectus carinatum appeared to be less satisfied with their appearance than those with pectus excavatum (p = 0.07). Body image distress was multivariately associated with both reduced mental quality of life and low self-esteem (p < 0.001). Body image did not influence physical quality of life. Patients displayed no elevated rates of mental disorders according to Diagnostic and Statistical Manual of Mental Disorders, fourth edition (DSM-IV) criteria. Conclusion: Since self-perception is a major contributor to therapeutic decision making, a systematic evaluation of body image should be included in the assessment of patients with chest deformities. Body image concerns may be even more relevant to the decision-making process than physical restrictions. Exaggerated dysmorphic concerns should be prospectively investigated in their ability to influence the extent of satisfaction with the surgical outcome.

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Keywords: Pectus excavatum; Pectus carinatum; Quality of life; Body image

## 1. Introduction

Pectus excavatum (PE) and pectus carinatum (PC) are the most common anterior chest-wall deformities. Both of these malformations predominantly affect males. They often coincide with the vulnerable life phase of puberty, a period characterized by great physical, social, and emotional changes. PE either can be present at birth or may develop and deteriorate during childhood and adolescence; PC usually manifests itself at the time of a growth spurt in the early teenage years. PE appears in as many as 1 in 400 to 1 in 1000 live births with a four to one male:female ratio; PC occurs less frequently [1].

Outer appearance becomes a major issue for adolescents, and those who display a visible disfigurement may be put at a disadvantage, having to come to terms with a difference in their looks and having to face the risk of harassment by their peers. Patients with pectus deformities often experience feelings of shame and try to hide their chests. This can be reflected in the choice of clothing or poor body posture. Many patients even avoid social activities and sports [2,3].

Chest-wall malformations can have a substantial impact on social interaction as well [2,4]; for example, quite a few patients experience stigmatization [2] and, at the same time,

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Table 1. Sociodemographic characteristics of control and patient groups.

|                        | CG                               | PE                               | PC                               | p value |
|------------------------|----------------------------------|----------------------------------|----------------------------------|---------|
| N                      | 82                               | 71                               | 19                               |         |
| Age (mean $\pm$ SD)    | $\textbf{20.4} \pm \textbf{5.6}$ | $\textbf{19.5} \pm \textbf{4.8}$ | $\textbf{19.3} \pm \textbf{5.1}$ | NS      |
| Education (N [%])      |                                  |                                  |                                  |         |
| School/university      | 49 (59.8)                        | 39 (54.9)                        | 12 (66.7)                        | NS      |
| Apprenticeship/working | 27 (32.9)                        | 20 (28.2)                        | 5 (27.8)                         |         |
| Other                  | 6 (7.3)                          | 12 (16.9)                        | 1 (5.6)                          |         |

NS: not significant; CG: control group; PE: pectus excavatum; and PC: pectus carinatum.

they may display inadequate social behavior or even avoid social situations [3]. As they mature, patients become increasingly concerned about sexuality and intimacy. They are aware of the fact that their deformity is generally not considered attractive, and some regard it as an obstacle in future relationships [2]. Reduced self-confidence and selfesteem appear to be rather common characteristics of PE patients [3,4].

Recently, several authors have assessed the improvement of quality of life (QoL) and body image following surgical repair of PE [2,5–11]. Yet, little is known about the degree to which patients, prior to surgery, differ from healthy individuals within the same age group. So far, only few studies have attempted to describe psychological strain in PE patients prior to corrective surgery [2–4,12]. Even less is known about possible differences in the impact of PE and PC on health-related QoL and body image. PC is the second most frequent chest-wall malformation. These patients are usually excluded from research. The few reports available are mainly concerned with overall satisfaction with the surgical outcome.

The aim of this study was to investigate the effects of chest deformities on mental well-being in patients with PE or PC, compared to a control group (CG). We focused on the evaluation of disease-specific and health-related QoL and body image and on the possible existence of symptoms for mental disorders. The second objective was to identify predictors of restrictions in physical and mental QoL and self-esteem.

#### 2. Material and methods

#### 2.1. Subjects

A total of 90 patients who were scheduled for surgical correction of PE or PC were recruited at the Department of Pediatric Surgery at the University Hospital of Erlangen, Germany. Patients were eligible for participation if they were male and aged between 14 and 40 years. Seventy-one PE patients with an age range of 14-35 years and 19 PC patients with an age range of 14-32 years fulfilled the inclusion criteria. To create a more homogeneous study population and to simplify the interpretation of the results, female subjects were excluded from recruitment.

Male controls were recruited via announcements in public places and were matched for age and education. This CG consisted of 82 individuals aged between 14 and 33 years and not affected by an anterior chest-wall deformity. As shown in Table 1, patients and control subjects did not differ in sociodemographic aspects.

## 2.2. Procedure

All participants were recruited between April 2007 and May 2010. Participation was voluntary and written informed consent was obtained from each subject. The study design was approved by the institutional review board and ethics committee of the Friedrich-Alexander University of Erlangen-Nürnberg.

Prior to surgery, patients had their medical history taken and were given a complete physical examination. On admission to hospital, patients were evaluated with a structured clinical interview, and they completed a generic questionnaire on impairment attributable to the chest malformation and on motivational reasons to undergo surgery. All patients and control subjects answered a sociodemographic questionnaire, including questions on educational and familial background. They also filled in several self-report questionnaires.

#### 2.2.1. Physical examination

During preoperative physical examination, the type of deformity (PE or PC), physical symptoms associated with the deformity, and posture problems were assessed. Additionally, the funnel-chest index (FCI) by Hümmer [13] was obtained as a quantitative indicator of the severity of the chest-wall deformity. The FCI is a non-invasive external method of objective evaluation that does not expose the patient to radiation. It takes into consideration the position of the sternum in relation to the vertebral column with focus on the minimal (PE) or maximal (PC) sagittal thoracic diameter. Values below the normal range of 115-145% are typically found in PE patients, while values above the norm usually pertain to PC patients [13]. For better use in the ensuing analyses, we created a 'severity index' constituting the positive deviance of each individual patient's FCI value from the medial norm (130%). In this severity index, higher values indicate a more pronounced pectus deformity. The FCI was chosen as a presumably preferential index for assessing the impact of outward appearance. To allow for better comparability of the present data with other reports, we also used computed tomography (CT) scans to calculate the Haller index from the internal thoracic transversal and anteroposterior diameters.

## 2.2.2. Diagnostic interview

The standardized Short Diagnostic Interview for Mental Disorders (Diagnostisches Kurzinterview bei psychischen Störungen, Mini-DIPS) [14] was administered to screen for mental disorders according to the Diagnostic and Statistical Manual of Mental Disorders, fourth edition (DSM-IV). The main diagnostic categories are as follows: anxiety disorder, obsessive—compulsive disorder, affective disorder, somatoform disorder, eating disorder, substance abuse, and psychotic disorder.

## 2.2.3. Self-report questionnaires

2.2.3.1. Quality of life. The Short-Form-36 Health Survey (SF-36) [15] is a multidimensional instrument to assess health-related QoL. It comprises two summary subscales: (1) the physical component summary (PCS) features aspects of 'physical functioning', 'physical role functioning', 'bodily pain', and 'health in general' and (2) the mental component summary (MCS) is based on the dimensions 'social functioning', 'emotional role functioning', 'vitality', and 'mental health'. Higher scores are indicative of better health.

The Nuss Questionnaire modified for Adults (NQ-mA) [8] assesses PE-specific QoL in terms of physical and psychosocial functioning and pre- and postoperative satisfaction. Restricted QoL and reduced overall satisfaction are reflected in decreased sum scores. The NQ-mA is a slightly altered version of the Pectus Excavatum Evaluation Questionnaire (PEEQ) [10]. Exploratory factor analysis using varimax rotation resulted in two factors, explaining 69.5% of variance: factor 1 (explaining 57.2% of variance) covers nine items related to body image and factor 2 (explaining 12.3% of variance) includes three items pertaining to physical difficulties attributable to the chest malformation. Recently, the PEEQ has been used with a similar factor structure [8].

2.2.3.2. Body image. The Body Image Questionnaire (Fragebogen zum Körperbild, FKB-20) [16] is a German 20-item assessment tool measuring body image disturbances based on the dimensions 'perception of body dynamics' (FKB-20-PBD) and 'negative evaluation of the body' (FKB-20-NEB). The first subscale focuses on perceived energy, fitness, and health. The second scale considers the satisfaction with one's physical appearance. Answers are given on a Likert scale ranging from 1 (not applicable) to 5 (fully applicable). Low FKB-20-PBD scores and high FKB-20-NEB scores suggest body image disturbance.

The Dysmorphic Concern Questionnaire (DCQ) [17] is a 7item questionnaire assessing concern about physical appearance covering affective, cognitive, and behavioral aspects. All items are rated on a 4-point Likert scale from 0 (not at all) to 3 (much more than most people). Elevated scores imply increased dysmorphic concern.

In addition, participants were asked to evaluate their physical appearance on a self-rating scale ranging from 0 (not at all impaired) to 10 (most severely impaired) [18]. Higher values reflect a higher degree of perceived impairment in physical appearance.

2.2.3.3. Other psychological indicators. Rating of selfesteem was conducted on a numeric rating scale ranging from 1 (low) to 10 (high) [8]. Higher values indicate better self-esteem.

The General Depression Scale (Allgemeine Depressionsskala, ADS) [19] is the German version of the Center for Epidemiological Studies – Depression Scale (CES-D). It consists of 20 items pertaining to typical current depressive symptoms. On a scale ranging from 0 (rarely or none of the time – less than 1 day) to 3 (most or all of the time – 5–7 days), respondents indicate how often they have experienced each symptom during the past week.

## 2.3. Statistical analysis

Comparisons between the two clinical groups (PE and PC) were done by using Student's *t*-test for parametric and  $\chi^2$  analysis for nonparametric data. In comparing the three study samples (PE, PC, and CG), we applied  $\chi^2$  analyses for

Table 2. Morphologic, physical, and psychosocial findings in pectus excavatum and pectus carinatum.

|   | PE                                  | PC           |           | <i>p</i> value |
|---|-------------------------------------|--------------|-----------|----------------|
|   | Mean $\pm$ SD                       | Mean $\pm$ S | D         | <i>t</i> -test |
| Funnel-chest index (%)                        | $\textbf{106.4} \pm \textbf{14.4}$  | $146.2\pm1$  | 0.000     |                |
| Severity index (Hümmer) (%)                   | $\textbf{24.5} \pm \textbf{12.9}$   | $19.6\pm1$   | 2.6       | NS             |
| Haller index                                  | $\textbf{4.4} \pm \textbf{1.5}$     | $2.0\pm0$    | 0.3       | 0.000          |
| Severity index (Haller)                       | $\textbf{1.8} \pm \textbf{1.5}$     | $0.6\pm0$    | ).3       | 0.000          |
|   |                                     | PE           | PC        | p value        |
|   |                                     | N (%)        | N (%)     | $\chi^2$ -test |
| Physical symptoms                             |                                     |              |           |                |
| Cardiopulmonary: reduced exercise endurar     | nce, dyspnea, palpitations, vertigo | 58 (96.7)    | 5 (33.3)  | 0.000          |
| Chest discomfort: chest tightness, thoracic   | pain                                | 48 (82.8)    | 7 (43.8)  | 0.002          |
| Postural: back pain, muscle tension           |                                     | 34 (58.6)    | 11 (61.1) | NS             |
| Gastro-esophageal: dysphagia, reflux          |                                     | 19 (32.8)    | 3 (20.0)  | NS             |
| Posture                                       |                                     |              |           |                |
| Flaring ribs                                  |                                     | 41 (66.1)    | 5 (29.4)  | 0.007          |
| Scoliosis, kyphosis                           |                                     | 46 (79.3)    | 14 (77.8) | NS             |
| Shoulder elevation, pelvic obliquity, scapula | ae alatae                           | 35 (62.5)    | 11 (64.7) | NS             |
| Psychosocial symptoms: Impairment of activit  | ies                                 |              |           |                |
| Overall                                       |                                     | 52 (73.2)    | 14 (73.7) | NS             |
| <ul> <li>School/workplace</li> </ul>          |                                     | 7 (10.1)     | 4 (21.1)  | NS             |
| <ul> <li>Physical exercise</li> </ul>         |                                     | 45 (63.4)    | 10 (52.6) | NS             |
| • Social                                      | 15 (21.4)                           | 10 (52.6)    | 0.007     |                |
| Relationship                                  |                                     | 24 (34.3)    | 10 (52.6) | NS             |
| <ul> <li>Sexual contact</li> </ul>            |                                     | 13 (18.6)    | 6 (31.6)  | NS             |

NS: not significant; PE: pectus excavatum; and PC: pectus carinatum.

categorical variables and analyses of variance (ANOVAs) for parametric data. To protect against inflated type I error rates, we conducted multivariate ANOVAs first in constructs consisting of multiple variables (disease-specific QoL via two subscales, health-related QoL, and body image), followed by the Welch test for univariate testing. This procedure similar to ANOVA does not require equal variances and is more conservative. If the overall tests were significant, they were followed by Games—Howell *post hoc* paired comparisons. The Games—Howell test does not assume homogeneity of variances or normal distribution and is suitable for unequal sample sizes. Finally, Cohen's *d* effect sizes were calculated as a standardized measure to determine the magnitude of differences in paired comparisons.

Controlling for the influence of chest configuration (PE or PC), we performed multiple regression analyses. Criterion variables were mental (MCS) and physical (PCS) QoL and selfesteem. Physical aspects of the deformity and body image scales were included as independent variables.

All tests were two-tailed with statistical significance being accepted at p < 0.05. All analyses were done with PASW Statistics (version 18, Chicago, IL, USA).

## 3. Results

## 3.1. Clinical characteristics of PE and PC

As shown in Table 2, the FCI and the Haller index were highly correlated (r = -0.68; p = 0.000). Table 2 also presents the percentage of subjects affected by various physical and psychosocial restrictions. During preoperative history taking, almost every PE patient reported physical symptoms. In

79.0% of PE patients, at least three different physical complaints were present. Only 3.2% of PE patients displayed no symptoms. In the PC group, 27.8% of the patients had no symptoms and only 44.4% reported three or more complaints. The mean number of physical symptoms was 4.5 ((95% confidence interval (CI): 4.0, 5.1), Md = 4.5) in PE patients and significantly lower in PC patients, who only displayed an average of 2.2 ((95% CI: 1.2, 3.1), Md = 2.0) symptoms, t(78) = 4.0, p < 0.001. PC patients were significantly less affected by cardiopulmonary restrictions and chest discomfort. Poor body posture was observed in 95.2% of PE and 94.4% of PC subjects. There was no significant difference in the average number of posture problems: 2.4 ((95% CI: 2.1, 2.7), Md = 2.0) in PE and 2.1 ((95% CI: 1.5, 2.6), Md = 2.0) in PC. Flaring ribs were observed less often in PC patients.

More than 70% of patients stated in the generic questionnaire that their chest anomaly impeded their daily activities. Social activities were more frequently impaired in PC than in PE patients.

Patients were asked for internal and external motivational factors to undergo surgery. There was a statistically significant difference in motivational reasons for corrective surgery,  $\chi^2(3) = 28.4$ ; p = 0.000. A combination of both cosmetic and physical health-related reasons was the main reason for 50.7% of PE patients. In 11.3% of PE patients, the cosmetic aspect alone was the primary reason for their decision. Physical health alone was the main argument in 23.9% of PE patients. In PC, 26.3% of the patients had both cosmetic and health-related reasons to undergo surgery. Another 68.4% of PC patients sought surgical correction for solely cosmetic reasons. No PC patient wanted surgery for physical health-related reasons only. The remaining participants (PE: 14.1%; PC: 5.3%) did not specify their reasons to undergo surgery.

Table 3. Between-group comparisons of disease-specific and health-related quality of life and body image.

| Variables                                    | CG   | CG  |      | PE  |      |     | Between-subject effects            | Post hoc pairwise comparison |                 |                 |
|--|------|-----|------|-----|------|-----|------------------------------------|------------------------------|-----------------|-----------------|
|  | м    | SD  | м    | SD  | м    | SD  | MANOVA/Welch's<br>test F (df1;df2) | CG versus<br>PE              | CG versus<br>PC | PE versus<br>PC |
| Disease-specific quality of life (NQ-m       | A)   |     |      |     |      |     |                                    |                              |                 |                 |
| Total score                                  | 44.0 | 4.3 | 30.2 | 5.4 | 30.1 | 5.6 | 163.6 (2;48.2) <sup>***, a</sup>   | ***                          | ***             |                 |
| NQ-mA scales                                 |      |     |      |     |      |     | 73.8 (4;328) <sup>***, b</sup>     |                              |                 |                 |
| Body image                                   | 32.3 | 3.9 | 21.0 | 5.1 | 19.1 | 5.1 | 139.5 (2;48.1) <sup>***, a</sup>   | ***                          | ***             |                 |
| Physical                                     | 11.6 | 1.0 | 9.3  | 1.8 | 11.1 | 1.0 | 48.2 (2;51.9) <sup>***, a</sup>    | ***                          | +               | ***             |
| Health-related quality of life (SF-36)       |      |     |      |     |      |     | 4.6 (4;330) <sup>***, b</sup>      |                              |                 |                 |
| Physical component summary                   | 55.3 | 5.7 | 52.5 | 6.1 | 55.1 | 5.4 | 4.2 (2;49.8) <sup>*, a</sup>       | *                            |                 |                 |
| Mental component summary                     | 48.9 | 9.0 | 46.6 | 9.3 | 42.4 | 8.7 | 4.2 (2;48.9) <sup>*, a</sup>       |                              | *               |                 |
| Body image                                   |      |     |      |     |      |     | 15.36 (8;312)*** <sup>, b</sup>    |                              |                 |                 |
| Dysmorphic concerns (DCQ)                    | 3.8  | 3.3 | 8.3  | 3.6 | 9.2  | 3.3 | 39.3 (2;49.1) <sup>***, a</sup>    | ***                          | ***             |                 |
| Rating of appearance                         | 1.9  | 2.0 | 5.4  | 2.6 | 6.2  | 1.9 | 64.1 (2;50.7) <sup>***, a</sup>    | ***                          | ***             |                 |
| Negative evaluation of the body (FKB-20-NEB) | 18.1 | 6.7 | 25.9 | 7.1 | 30.2 | 7.2 | 35.4 (2;50.2) <sup>***, a</sup>    | ***                          | ***             | +               |
| Perception of body dynamics<br>(FKB-20-PBD)  | 37.9 | 4.7 | 32.9 | 5.0 | 31.9 | 5.6 | 23.3 (2;48.9) <sup>***, a</sup>    | ***                          | **              |                 |

CG: control group; PE: pectus excavatum; PC: pectus carinatum; DCQ: Dysmorphic Concern Questionnaire; FKB-20-NEB: negative evaluation of the body – subscale of the Body Image Questionnaire; FKB-20-PBD: perception of body dynamics – subscale of the Body Image Questionnaire; NQ-mA: Nuss Questionnaire modified for Adults; and SF-36: Short-Form-36 Health Survey.

<sup>a</sup> Welch's test results.

<sup>b</sup> MANOVA results.

\* *p* < 0.05.

*p* < 0.01.

*p* < 0.001.

<sup>+</sup> *p* < 0.10.

| Variables                    | CG   |     | PE   | PE  |      |     | Between-subject effects     | Post hoc pairwise comparison |                 |                 |
|------------------------------|------|-----|------|-----|------|-----|-----------------------------|------------------------------|-----------------|-----------------|
|                              | м    | SD  | М    | SD  | м    | SD  | Welch's test<br>F (df1;df2) | CG versus<br>PE              | CG versus<br>PC | PE versus<br>PC |
| Depressive symptoms<br>(ADS) | 12.2 | 8.0 | 11.3 | 8.2 | 14.5 | 6.5 | 1.6 (2;55.4)                |                              |                 |                 |
| Self-esteem                  | 7.9  | 1.5 | 6.8  | 1.6 | 6.0  | 2.2 | 14.0 (2;41.6)***            | ***                          | **              |                 |

Table 4. Between-group comparisons of depressive symptoms and self-esteem.

CG: control group; PE: pectus excavatum; PC: pectus carinatum; and ADS: General Depression Scale.

\*\* *p* < 0.01.

p < 0.01.

## 3.2. Quality of life

PE, PC, and CG were compared with regard to differences in various aspects of QoL. A significant group difference was obtained for disease-specific QoL, as indicated by the NQ-mA sum score (see Table 3). Mean scores were markedly reduced in patients, compared to those of the CG. Highly significant main effects resulted from multivariate analysis of variance (MANOVA) of the individual NQ-mA subscales (body image and physical difficulties), Wilk's  $\lambda = 0.28$ , F(4, 328) = 73.84, p = 0.000, and from univariate testing of these subscales. Both the patient groups displayed a higher degree of body image impairment than the CG. Cohen's d revealed large effect sizes (d = 2.5 for CG vs PE; d = 2.9 for CG vs PC). Compared to those in control subjects, physical difficulties were significantly increased in PE patients (Cohen's d = 1.6), but there was only a trend for significance in PC patients (p = 0.096).

There was a significant main effect of MANOVA for general health-related QoL (SF-36), Wilk's  $\lambda = 0.90$ , F(4, 330) = 4.61, p = 0.001. Univariate testing revealed significant main effects for physical and mental QoL subscales as well. In a pairwise comparison, PE patients attained significantly lower PCS scores than the control subjects (Cohen's d = 0.5). Their MCS scores did not differ from CG results. By contrast, MCS scores were significantly reduced in PC patients (Cohen's d = 0.8), while PCS scores were equal to control results.

#### 3.3. Body image

As stated earlier, the body-image-related NQ-mA subscale showed a higher degree of body image problems in patients with a chest malformation. Patients scored worse than control subjects not only on the disorder-specific assessment instrument, but also on every instrument assessing body image in general. MANOVA of non-disease-specific body image variables (DCQ, self-rating of appearance, FKB-20-NEB, and FKB-20-PBD) revealed a highly significant main effect for chest configuration, Wilk's  $\lambda = 0.52$ , *F*(8, 312) = 15.36, p = 0.000. Univariate analyses showed highly significant main effects for each of these body image variables. Highly significant pairwise differences were obtained from CG versus PE and from CG versus PC. All comparisons of clinical groups with the CG showed large effect sizes (1.0  $\leq d \leq$  2.2). Pairwise testing of PC versus PE patients revealed a trend toward a higher degree of body image impairment in PC patients as indicated by higher FKB-20-NEB scores (p = 0.07).

#### 3.4. Prevalence of comorbid mental disorders

Neither PE nor PC was associated with increased rates of mental disorders according to DSM-IV criteria assessed by the structured clinical interview (Mini-DIPS). The groups did not differ significantly with regard to current and past prevalence rates of psychiatric disorders. In 14.3% of PE (N = 10) and 5.3% (N = 1) of PC, patients met full criteria for a current diagnosis of mental disorder (PE: 2.9% affective disorder, 1.4% affective disorder with social phobia, 5.7% specific phobia, 1.4% substance abuse, 1.4% obsessive-compulsive disorder, and 1.4% somatoform disorder; PC: 5.3% affective disorder). In 16.9% (N = 12) of PE and 15.8% (N = 3) of PC, patients tested positive for any mental disorder in the past (PE: 14.3% affective disorder, 1.4% specific phobia 1.4% attention deficit hyperactivity disorder (ADHD); PC: 10.5% affective disorder, 5.3% social phobia). No eating or psychotic disorders were observed. As shown in Table 4, mean ADS scores did not differ between patients and control subjects, indicating no elevated rates of depressive symptoms in both clinical groups. However, selfesteem was markedly lower in pectus patients.

## 3.5. Supplementary analyses

The Shapiro-Wilk test showed deviation from normal distribution mainly in the CG. The reason for this is that in the tests we used a healthy person would score either very high or very low, depending on the specific questions. As a consequence, we performed additional analyses to further validate our findings. Overall, nonparametric testing revealed a very comparable pattern of results showing reduced QoL and increased body image disturbance in pectus patients as compared with controls. Only the following results were found to be different: Kruskal-Wallis was significant for the ADS score (p = 0.034). At the Bonferronicorrected  $\alpha$  level of p = 0.017, the ensuing Mann–Whitney U tests revealed a higher degree of depressiveness for PC, compared to PE (p = 0.015), but not compared with controls. PC scored significantly lower than the CG in the physical subscale of the NQ-mA (p = 0.000).

## 3.6. Associated features of health-related QoL and selfesteem

We used multiple regression analyses to identify predictors of mental and physical health and self-esteem in participants with a chest-wall anomaly. Independent variables were entered blockwise. The first block contained the severity of physical aspects associated with the deformity.

<sup>&</sup>lt;sup>\*</sup>p < 0.05.

| Table 5. Multiple regression | analyses of menta | l and physical health and | self-esteem in pectus patients. |
|------------------------------|-------------------|---------------------------|---------------------------------|
|                              |                   |                           |                                 |

|  | Mental component<br>summary (SF-36) |              | Physical component<br>summary (SF-36) |              | Self-esteem |              |
|--|-------------------------------------|--------------|---------------------------------------|--------------|-------------|--------------|
|  | β                                   | $\Delta R^2$ | β                                     | $\Delta R^2$ | β           | $\Delta R^2$ |
| 1  |                                     |              |                                       |              |             |              |
| Physical aspects                             |                                     | .08          |                                       | .20**        |             | .06          |
| Type of deformity (PE or PC)                 | 06                                  |              | .07                                   |              | 05          |              |
| Severity index                               | 04                                  |              | 02                                    |              | 03          |              |
| Number of physical complaints                | .01                                 |              | 38**                                  |              | 12          |              |
| Number of posture problems                   | .10                                 |              | 03                                    |              | 08          |              |
| 2  |                                     |              |                                       |              |             |              |
| Body image                                   |                                     | .29***       |                                       | .04          |             | .42***       |
| Dysmorphic Concern Questionnaire (DCQ)       | .04                                 |              | 13                                    |              | .03         |              |
| Negative Evaluation of the Body (FKB-20-NEB) | 44**                                |              | 20                                    |              | 64***       |              |
| Body image scale (NQ-mA)                     | .18                                 |              | 36                                    |              | .09         |              |
| Total R <sup>2</sup>                         |                                     | .37***       |                                       | .23*         |             | .48***       |

PE: pectus excavatum; PC: pectus carinatum; FKB-20-NEB: negative evaluation of the body – subscale of the Body Image Questionnaire; NQ-mA: Nuss Questionnaire modified for Adults; and SF-36: Short-Form-36 Health Survey.

<sup>\*\*</sup> *p* < 0.01.

<sup>\*\*\*</sup> *p* < 0.001.

Body image variables were entered into the second block. Table 5 shows a complete summary of the results.

The type and severity of the deformity had very little impact on each regression model.

Mental health, as a criterion variable, was significantly associated with the FKB-20 subscale 'negative evaluation of the body' (FKB-20-NEB). Body image variables as a whole contributed to a significant rise of 29% of the explained variance of mental health. Physical aspects made no significant contribution. The final regression model accounted for 37% of explained variance.

The number of physical complaints showed to be the only significant predictor of QoL related to physical health. Physical aspects as a whole explained 20% of variance in PCS scores. Body image concerns were not related to physical health scores, with the incremental  $R^2$  not being significant. The final regression model explained 23% of variance.

Physical variables were not associated with self-esteem, but there was a strong association of body image variables with self-esteem. 'Negative evaluation of the body' was the most influential single variable. Body image variables as a whole accounted for 42% of explained variance, the total explained variance being 48%.

Taken as a whole, a 'negative evaluation of the body' was the most influential predictor of mental health-related QoL and self-esteem. Physical aspects of the chest malformation were mainly relevant to physical health-related QoL.

Performing these regression analyses with a severity index derived from the Haller index instead of the one derived from the FCI led to similar results (data not reported).

## 4. Discussion

We observed reduced disease-specific and general healthrelated QoL in patients with a chest malformation. Body image was highly impaired in both PE and PC patients. Body image dissatisfaction and, most notably, negative evaluation of the body, was a major predictor of restrictions in QoL and self-esteem. These findings of lower QoL and higher body image distress are in line with reports of QoL and body image improving with corrective surgery of PE [2,5–11].

In accordance with previous findings [1], we observed far fewer cardiopulmonary symptoms in PC patients than in PE patients. In the scale measuring physical health-related QoL, PC patients' scores were almost equal to the CG results. PE patients attained significantly lower scores than control subjects. By contrast, mental health-related QoL was reduced in PC patients, but not in PE patients.

Results suggest that PC patients might have not only more body image concerns than control subjects, but also more than PE patients. PC patients not only scored worse than PE patients on the mental QoL subscale, but also reached slightly lower scores in all body image questionnaires. One outcome measure, negative evaluation of the body, showed a trend toward significance between PE and PC. PC patients also reported significantly more restrictions in their social activities. We therefore suspect higher levels of psychological distress, possibly associated with PC. Considering the difficulty of hiding the chest-wall protrusion beneath one's clothing, as it remains visible even when loose attire is worn, PC patients seem to be at risk for increased mental distress [1]. Social withdrawal due to feelings of shame or even due to bullying experiences might especially present a problem to younger patients.

Previous observations comparing patients with facial and truncal acne vulgaris have found better self-esteem and body image in patients whose disorder was strictly limited to the trunk and therefore not visible to others when concealed beneath the clothing [20]. However, another study has shown that the visibility of burn scars has a significant, but only lowto-moderate, effect on body esteem, as opposed to certain social and emotional factors that are far more influential [21]. In conclusion, differences in psychological distress observed between PE and PC patients might, at least to a certain extent, be attributable to social or emotional factors. Nevertheless, considering that 94.7% of PC patients and only 62.0% of PE patients specified cosmetic reasons as a major argument for the decision to undergo corrective surgery, the visibility of the deformity is likely to be of particular

<sup>\*</sup> *p* < 0.05.

relevance. However, the relatively small number of PC patients in our sample does not allow for a concise conclusion. Therefore, future research on chest deformities should further investigate possible psychosocial effects resulting from PC.

Cosmetic impairment was important to PE patients, too, but to a lower degree than it was to PC patients. Unlike PC patients, the vast majority of PE patients in the present sample reported physical complaints associated with the chest-wall disorder. Nevertheless, cosmetic reasons to undergo surgical correction were almost equal in their importance as were physical health-related reasons. This result is consistent with previous findings [2].

Many surgeons are concerned about the possible existence of underlying psychological problems in patients, which may, in turn, hinder a successful chest correction. This notion contributed to the design of this study. So far, we have not observed any elevated rates of psychiatric disorders in preoperative patients. The prevalence of mental disorders in our clinical sample did not differ from the rates found in German adolescents and young adults [22]. Of course, there still exists the possibility of psychological symptoms that develop or aggravate during follow-up. On the whole, the central aspect of psychological distress associated with an anterior chest-wall malformation appears to be body image distress. We observed high rates of decreased body esteem in patients presenting themselves for corrective surgery, which became apparent in every individual scale assessing aspects of body image. Body image dissatisfaction was associated with reduced mental health-related QoL. This supports previous findings of body image having a major impact on QoL. Areas negatively affected by body image distress among others include behavioral constraint, situational avoidance, physical exercise, day-to-day emotions, social interaction with the other sex, and confidence [23]. These domains are frequently impaired among patients suffering from PE [2-4,10,12]. We also detected a strong multivariate association of body image distress with lower levels of self-esteem, yet another typical characteristic of PE [2,3]. This link between body image and self-esteem has already been observed in healthy individuals [24].

Patients' high degree of body image dissatisfaction resulted in reduced QoL and impaired self-esteem. However, it is important to bear in mind that body image may affect other areas of mental well-being as well, even to a lifethreatening degree in more severe cases. Negative body attitudes and feelings have been shown to have a stronger influence on suicidal ideation than depression, hopelessness, and past suicidal behavior [25].

Many reports about psychological health in pectus patients examine largely pediatric or adolescent collectives [2-5,7,9-12]. The present study sample consists of both adolescent and adult patients, ranging from 14 to 35 years of age. We therefore controlled for age to ascertain that our results are comparable with previous reports. Age did not appear to considerably influence mental well-being in our patients (data not reported).

We recognize the possibility of selection bias due to our CG being self-referred and therefore not necessarily representative of the general population. However, a descriptive comparison of CG scores with the normative values available

from the test manuals of SF-36, FKB-20, and ADS did not suggest a consistent deviation from the norm. CG results showed a good agreement with normative scores.

The present findings are the first results of a longitudinal prospective survey following patients undergoing corrective surgery for PE and PC, focusing on changes in QoL, body image, and other psychological variables. To our knowledge, this study is the first to compare pectus patients with a CG prior to surgical correction. Another strength of this study is that it employs standardized and non-disease-specific assessment tools. In previous studies, comparisons with normative values or CG results have rarely and — if at all — only postoperatively been implemented [5,9].

The PEEQ and its equivalent for older patients, the NQmA, are well-established assessment instruments. They have been specifically created for patients suffering from PE; comparative data from a healthy normative population do not exist, which makes it difficult to evaluate psychological impairment in relation to individuals not affected by a chestwall disorder. Up until now, only one group of researchers has assessed PE patients with a set of non-disease-specific standardized psychometric tests prior to surgery — but mainly as part of a survey which focused on postoperative changes [6].

There is an astounding lack of research on psychological health in patients with PC. Our study is the first to address psychological health in PC patients and to explore possible differences in psychological distress between PE and PC.

However, our conclusions are limited as a result of the relatively small sample of PC patients. Apart from that, in order to achieve a more homogeneous study population and to avoid potential confounding by gender-related anatomical differences, we excluded female patients from recruitment. Finally, and importantly, female subjects, as compared to male subjects, tend to display a higher degree of psychological distress, for example, a more negative body image [16,23] or reduced levels of QoL [15]. As a consequence, our results might only partially be applicable to female pectus patients. Moreover, we included only those patients who were already listed for surgical correction at the time they agreed to participate in this study. It stands to reason that our findings may not apply to patients who decide against surgery. Possible reasons for their decision might be lack of physical symptoms, lower degrees of psychological strain, as well as better coping strategies. Reasons as to why some individuals with an anterior chest-wall deformity of similar severity do not request corrective surgery are not yet fully identified and further research is needed. Even less is known about those individuals who never seek medical consultation in the first place.

In conclusion, systematic evaluation of body image should become a diagnostic standard in the assessment of patients presenting themselves for correction of an anterior chestwall malformation. A negative perception of the body considerably diminishes QoL, frequently even outweighing physical factors. Therefore, future research should continue to focus on body image. First, to determine whether or not surgical intervention is a sufficient measure to restore body image to levels found in the general population. Second, to examine how dysmorphic concerns can interfere with patient satisfaction after surgical repair.

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