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# The Outcome of the Single Step Questionnaire in Pectus Excavatum Patients is Phase Dependent

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Abstract Introduction Pectus excavatum (PE) is the most common chest wall deformity. Patients with PE may have cosmetic complaints, restricted physical capabilities, or both and may seek surgical correction. One method to assess satisfaction after surgery is the single step questionnaire (SSQ). Although the developers state that the SSQ produces a stabile score and only needs to be used once, we hypothesized that the score may depend on point in time after surgery. **Materials and Methods** One hundred and eight patients from a longitudinal cohort of patients undergoing a Nuss bar placement for PE were selected. Mean age was 16.0 years (range: 12-29). SSQ was completed at 6 weeks, 6 months, 1 year, and 2 years postoperatively. Mean and median scores per question and total scores were calculated on each measurement moment. Overall scores were tested using the Friedman test. **Results** There were significant differences in overall SSQ scores (p < 0.009) throughout the postoperative period, especially between 6 weeks and 6 months (p = 0.006). Scores on general health, exercise capacity, impact on social life, pain during hospital stay, and after discharge changed also significant in the first 2 years after Nuss bar **Keywords** placement. **Conclusion** There were significant differences in total SSQ score depending on the pectus time of application postoperatively. However, the most clinical relevant difference was pediatric thoracic between 6 weeks and 6 months. Assessment of the overall satisfaction postoperative with the SSQ questionnaire should not be done with a single measurement but rather at outcome different postoperative time intervals before and after 6 months postoperatively. surgery

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# Introduction

Pectus excavatum (PE) is a congenital malformation of the chest wall. It is the most common deformity of the chest wall and affects more males than females. Patients with PE may seek medical advice because of cosmetic and/or physical complaints. Surgical correction with the implantation of the Nuss bar is more and more widely used. It is a minimally invasive procedure by which the deformity of the chest wall is corrected with the use of a metal bar. The procedure is relative easily done; however, it is generally considered as a painful operation. To evaluate the satisfaction with this surgical technique, different questionnaires were developed. The single step questionnaire (SSQ) was developed by Krasopoulos et al in an attempt to provide a simpler assessment tool to measure satisfaction for young adults who underwent surgical correction of their PE with a Nuss bar. The tool addresses specific items associated with satisfaction but can also produce an overall score, whereby scores more than 41 (max 84) are considered as satisfactory result.<sup>1</sup>

The specific questions of the SSQ reflect changes on areas such as self-esteem, body image, social activities, pain, and cosmetics. The SSQ consists of 16 questions, but for the total score question 8 is subtracted from question 9 to form one score, thus adding up 15 scores to come to the total score. We believe the assessed areas may be subject to shifts over time, which could affect the SSQ value(s) and consequently making the scores of the SSQ depending on the moment of completion. This would imply that the SSQ is not a questionnaire that can be limited to one measurement. The aim of this study is to find out whether or not the results of the SSQ are time-dependent.

# **Materials and Methods**

The study is based on a longitudinal cohort of PE patients. This multicenter study cohort included 108 subjects (14 female, 94 male) with a mean age of 16 years (standard deviation [SD]: 2.20), treated with Nuss bar correction of their PE.

Patients who underwent PE surgery, above the age of 12 years, with implantation of a Nuss bar between 2011 and 2016 at one of five participating centers (AMC, VUMC, UMCG, Juliana Children's Hospital/Haga-Hospital, Radboud UMC) were asked to complete the SSQ postoperatively at 6 weeks (T1), 6 months (T2), and 12 months (T3). Patients with insufficient knowledge of the Dutch language in reading or writing were excluded. Patients with Marfan syndrome or other associated connective tissue diseases were allowed to participate.

All patients gave informed consent, as did their parents if they were younger than 16 years. The medical ethics committee approved the study.

The scores on individual questions of the SSQ were compared at 6 weeks, 6 months, 12 months, and 24 months postoperatively after correction of the PE. Next to that the overall score of the SSQ on all four moments in time was calculated, whereby the item self-esteem was calculated by subtracting the score to question 8 (preoperative selfesteem) from the score on question 9 (postoperative selfesteem).

In all study patients with PE, the Nuss procedure was performed.<sup>2</sup> Surgery was performed by dedicated pediatric or thoracic surgeons. The operative technical procedure was similar in all centers. Postoperative pain management was done using epidural analgesia or patient-controlled intravenous analgesia using morphine and occasionally ketamin. When possible, pain medication was changed after 3 days to oral medication.

#### **Statistical Analysis**

Data analyses were conducted using the IBM SPSS 23 software (SPSS Inc. Chicago, Illinois, United States). Descriptive statistics for variables are presented as means and SDs. The mean scores were calculated as were the median scores, to get a better assessment of the change of satisfaction in the separate items over time as compared with the scores of each item at different time points. Comparison between scores at measurement moment T1, T2, T3, and T4 for the enlisted variables from the study group were calculated using the Friedman test. Evaluation of the measurements at T1 versus T2 and T2 versus T3 and T3 versus T4 were performed using Wilcoxon signed-rank test. The cut-off point for significance was set at p-value of < 0.05.

# Results

Nighty-nine patients received one Nuss bar and nine a double Nuss bar implantation. Seventy-eight percent had one stabilizer per bar. Thirteen patients received two stabilizers on each side of their bar. Median admission time was 7 days (range: 4–16 days) of which one day was preoperative.

Patient received in 91% pre- and postoperative epidural analgesia. It took on average 3.9 days to change the epidural medication to oral medication. Patients with 2 bars needed on average 4.0 days. The complications during the first year postoperative were 7 cases of sensibility disorder while using epidural analgesia direct postoperative, which all resolved spontaneously. Peroperatively, the pericardium was opened in three and the diaphragm in one patient, which were all managed conservatively. Four patients were treated with antibiotics for a wound infection, urinary tract infection, pericarditis, and pneumonia, respectively. There were three reinterventions all of which concerned the stabilizer. In one case, a stabilizer was removed because of persistent pain, in one patient a stabilizer was added because of Nuss bar turn over, and in a third patient a stabilizer was readjusted.

Scores on "general health," "exercise capacity," and "impact on social life" changed significant in the first 2 years after Nuss bar implantation. Although pain experienced in daily life decreased over time, it remained a negative influence especially in the first 6 months, see **Tables 1** and **2**. This means that at 6 weeks, 18% of patients had a lot or a little bit of pain during activities, decreasing to 8% at 6 months and

SSO	6 weeks (T1)	6 months (T2)	1 vear (T3)	2 years (T4)	<i>p</i> -Value
Health in general after the operation	4	4	4	4	F
Exercise capacity after the operation	3	3	4	3	
Extent that chest looks interfere with preoperative social activity	3	3	3	3	
Extent that chest looks interfere with postoperative social activity	4	5	5	5	
Satisfaction with the overall postoperative appearance	4	4	4	4	
Bothered by the surgical scars	4	5	5	5	
Impact operation had to social life	3	4	4	4	
Preoperative self-esteem	6	6	6	6	
Postoperative self-esteem	8	8	8	8	
Pain during hospital stay	2	2	2	2	
Pain interfering with day-to-day activity now	3	4	4	4	
Pain now	4	5	5	5	
Conscious about the metallic bar	3	4	4	4	
Overall satisfaction with the final result	4	4	4	4	
Chest looks different	5	5	5	5	
Going back, would you have the operation again	10	10	10	10	
Total score SSQ	58	64	65	64	< 0.001

Table 1 Median SSQ scores over time
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Abbreviation: SSQ, single step questionnaire.

Note: The numbers are expressed in median. For testing of total score, the Friedman test was used for 4 paired samples of semicontinuous variable. Significance was set at p-value of < 0.05.

5% at 2 years. Painkillers were used after 6 months by 5%, and after 24 months by 3% of patients.

Most of the patients were satisfied with their scar 6 weeks after operation and remained satisfied afterwards.

The total scores of the SSQ in mean and median values were significantly different at 6 weeks, 6 months, 1 year, and 2 years after PE correction with a Nuss bar. Differences were especially noticeable between the 6 weeks and 6 months interval questionnaires, although the total score was increasing over the complete timeframe (see **– Table 2**). The patients who received implant of two bars had a lower total score of the SSQ at all four measurement moments although not significant (p = 0.181).

# Comment

In the literature, little is known about comparison of the SSQ scores at different time intervals. One study that gives a little information was performed by Metzelder et al who showed a persisting satisfaction after a mean period of 23 months after bar removal of the Nuss procedure, without significant differences in age groups or sex. They asked only a few questions of the SSQ after 6 months and took a one time measurement of the whole SSQ after a mean of 23 months.<sup>3,4</sup> In the study of Hanna et al with a median follow-up of 44.6 months, the SSQ showed a majority reporting improvement

in their appearance and well-being after the operation.<sup>5</sup> However, in general, studies measuring quality of life and body image show that these items are not stabile over time.<sup>6,7</sup> Considering the fact that self-esteem and body image are an important part of the SSQ score, the recommendation of the SSQ being a questionnaire that only needs to be applied once seemed doubtful.

To come to sound conclusions, the differences between the study set-up of Krasopoulos et al and our study were assessed. Since Krasopoulos et al only used a small group of 20 male patients, we also looked at our own results for both sexes apart.<sup>1</sup> The mean age of the male participants at the time of operations was 16.03 years (median: 15.51 years, range: 12–29). The female patients had a mean age of 15.58 years (median: 15.56 years, range: 12–20). The mean total score of the males were 57.16, 59.12, 60.13, and 60.10, respectively, at 6 weeks, 6 months, 1 year, and 2 years. For females, the mean total score was slightly lower with 55.43, 56.93, 57.04, and 58.75 at the same moments in time. This was not statistically significantly different (p = 0.161).

Pain was the most important item that influenced the satisfaction after Nuss bar placement, and thereby the total score of the SSQ, on the different measurement points in time. The SSQ pain scores postoperative were at an increased level at 6 weeks compared with preoperative and decreased afterwards.

Table 2 Mean SSQ scores over time

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SSQ	6 weeks (T1)	T1 versus T2, <i>p</i> -value	6 months (T2)	T2 versus T3, <i>p</i> -value	1 year (T3)	T3 versus T4, <i>p</i> -value	2 year (T4)	Overall <i>p</i> -value
Health in general after the operation	3.63 (1.00)	0.10	3.91 (0.99)	0.104	4.05 (0.89)	0.116	3.92 (0.84)	0.003
Exercise capacity after the operation	2.80 (1.20)	< 0.001	3.56 (1.11)	0.936	3.59 (1.08)	0.157	3.41 (1.06)	< 0.001
Extent that chest looks interfere with preo- perative social activity	2.81 (1.25)	0.154	2.66 (1.13)	0.186	2.80 (1.20)	0.988	2.88 (1.26)	0.188
Extent that chest looks interfere with post- operative social activity	3.94 (1.23)	0.029	4.24 (1.12)	0.971	4.21 (1.17)	0.962	4.26 (1.21)	0.269
Satisfaction with the overall postoperative appearance	3.97 (0.74)	0.773	3.99 (0.70)	0.539	4.03 (0.86)	0.687	4.11 (0.75)	0.234
Bothered by the surgical scars	4.20 (1.03)	0.903	4.23 (1.06)	0.259	4.34 (1.07)	0.363	4.14 (1.19)	0.449
Impact operation had to social life	3.49 (0.76)	0.354	3.57 (0.73)	0.085	3.69 (0.74)	0.262	3.74 (0.68)	0.013
Preoperative self-esteem	5.91 (1.661)	0.773	6.00 (1.62)	0.146	5.81 (1.62)	0.898	5.90 (1.51)	0.443
Postoperative self-esteem	8.14 (1.102)	0.144	8.00 (1.04)	0.344	7.94 (1.11)	0.560	8.07 (0.97)	0.432
Pain during hospital stay	2.23 (0.933)	0.464	2.19 (0.92)	0.648	2.16 (0.98)	0.211	2.12 (0.94)	0.033
Pain interfering with day-to-day activity now	3.42 (0.948)	< 0.001	4.01 (0.91)	0.945	4.02 (0.91)	0.310	4.11 (0.93)	< 0.001
Pain now	4.02 (1.014)	< 0.001	4.44 (0.87)	0.884	4.44 (0.78)	0.097	4.53 (0.69)	< 0.001
Conscious about the metallic bar	3.22 (1.113)	0.066	3.41 (1.02)	0.538	3.34 (1.15)	0.564	3.46 (1.15)	0.556
Overall satisfaction with the final result	4.09 (0.743)	0.649	4.06 (0.71)	0.873	4.05 (0.74)	0.303	4.01 (0.77)	0.671
Chest looks different	4.69 (0.541)	060.0	4.62 (0.58)	0.144	4.56 (0.60)	0.655	4.55 (0.60)	0.168
Going back, would you have the operation again	8.19 (3.092)	0.369	7.96 (3.28)	0.122	8.33 (3.21)	0.608	8.55 (2.92)	0.067
Total score SSQ	56.94 (7.84)	0.006	58.85 (8.37)	0.202	59.73 (8.61)	0.832	59.95 (7.53)	0.009
Abbreviation: SSQ, single step questionnaire.								

Notice and the preservation. Such a preservation of scores, the Friedman test was used for 4 paired samples of semicontinuous variable. The p-values of the paired samples of T1 versus T2 and T2 versus T3 and T3 versus T4 were calculated using the Wilcoxon signed-rank test. Significance was set at p-value of < 0.05.

Pain is also present in the normal nonsurgical adolescent school population. A study of Haraldstad et al showed that pain problems are highly prevalent in adolescents and health-related quality of life (HRQoL) scores are impaired in adolescents with pain compared with peers without pain. Girls were more affected than boys.<sup>8</sup> In addition to this "normal" pain, the PE patients suffer from pain due to the surgery. This pain persists long after the admission period and thus influences scores of any questionnaire that assesses pain. However, when the pain diminished, the influence of pain on the overall score changes implying that time of assessment can cause differences in total scores due to differences in pain scores.<sup>9</sup>

Furthermore, healthy school children show differences in HRQoL over time which seems merely influenced by age and sex.<sup>10</sup> One may expect that the answers to the SSQ questions regarding pain, but also self-esteem, will also be influenced by gender, age, physical, and psychological well-being, and differ in time independently of surgery.

Adolescence is a time of great changes physically as well as hormonal, social, psychological, and sexual. It could be argued that even in adolescents without PE responses on serial questionnaires would lead to different scores over time as a reflection of the phase the adolescent is in.<sup>11</sup> This could also have an effect on the outcome of the measurement of total score of the SSQ.

Patients were not really worried immediately after surgery by their scars and even bothered less after 6 months. Although cosmesis is the most important reason for engaging in surgery of the chest wall, it appears that an improvement is sought and not so much a perfect chest wall. So fitting into the group and having a "normal look" seems to be important in adolescence, where peer pressure is high.<sup>12</sup>

Since the study of Krasopoulos et al used the median scores for calculation of the SSQ, we calculated both the median scores as well as the little more specific mean scores of the SSQ. Our study group showed significant differences in overall SSQ scores regardless of the use of median or mean scores over the postoperative period of 6 weeks, 6 months, 1 year, and 2 years, with the largest differences being between 6 weeks and 6 months. Although the differences are statistically significant, the changes after 6 months postoperatively are small and probably not clinically significant. It would be interesting to see if the SSQ would reach a steady state after removal of the Nuss bar. This is usually done after approximately 3 years. However, since the patient groups undergoing Nuss bar implantation are in their adolescence, it could also be that to reach a steady state in SSQ total scores patients need to have left the adolescence period behind reaching adulthood with its phase of more psychological stability. It would be worthwhile to test this hypothesis in the near future.

### Conclusion

There are significant differences in total SSQ scores depending on the time of application postoperative. As long as it is not established that patients do reach a steady state after PE correction, we recommend that the SSQ should not be used as a single-step questionnaire but administered at different postoperative time intervals, especially before and after 6 months postoperatively, to come to a more exact assessment of the overall satisfaction postoperative.

#### **Conflict of Interest**

None declared.

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