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Chapter 7

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 stable 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 post-operatively. 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 post-operative 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.

Conclusions

There were significant differences in total SSQ score depending on the time of application post-operatively. However, the most clinical relevant difference was between 6 weeks and 6 months. Assessment of the overall satisfaction post-operative with the SSQ questionnaire should not be done with a single measurement but rather at different post-operative time intervals before and after 6 months post-operatively.

Introduction

Pectus excavatum 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 pectus excavatum 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. In order 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 in young adults who underwent surgical correction of their pectus excavatum (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 [1].

The specific questions of the SSQ reflect changes on areas 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.

Patients and Methods

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

Patients who underwent pectus excavatum 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 questionnaire post-operatively at 6 weeks (T1), 6 months (T2), 12 months (T3) and 24 months (T4). Patients with insufficient knowledge of the Dutch language in reading or writing were excluded. Patients with Marfan's 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 post-operatively after correction of the pectus excavatum. 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 (pre-operative self-esteem) from the score on question 9 (post-operative self-esteem).

In all study patients with PE the Nuss procedure was performed [2]. Surgery was carried out by dedicated pediatric or thoracic surgeons. The operative technical procedure was similar in all centers. Post-operative 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 IBM SPSS 23 software (SPSS Inc. Chicago, IL, USA). Descriptive statistics for variables are presented as means and SDs. The mean scores were calculated as were the median scores, in order to get a better assessment of the change of satisfaction in the separate items over time as compared to 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 $p < 0.05$.

Results

Ninety-nine patients received one Nuss bar and nine a double Nuss bar implantation. Seventy-eight percent had one stabilizer per bar. Thirteen patients received 2 stabilizers on each side of their bar. Median admission time was 7 days (range 4-16 days) of which one day was pre-operative.

Patients received in 91% per- and post-operative 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 post-operative were 7 cases of sensory disorder while using epidural analgesia direct postoperative, which all resolved spontaneously. Per-operatively 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 re-interventions 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 re-adjusted.

Scores on 'general health', 'exercise capacity' and 'impact on social life' changed significantly 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 Table 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 5% at two 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.

Table 1. Median scores SSQ over time

| SSQ | 6 weeks (T1) | 6 months (T2) | 1 year (T3) | 2 years (T4) | p-value |
|---|-----------------|------------------|----------------|-----------------|---------|
| Health in general after the operation | 4 | 4 | 4 | 4 | |
| Exercise capacity after the operation | 3 | 3 | 4 | 3 | |
| Extent that chest looks interfere with pre-operative social activity | 3 | 3 | 3 | 3 | |
| Extent that chest looks interfere with post-operative social activity | 4 | 5 | 5 | 5 | |
| Satisfaction with the overall post-operative appearance | 4 | 4 | 4 | 4 | |
| Bothered by the surgical scars | 4 | 5 | 5 | 5 | |
| Impact operation had to social life | 3 | 4 | 4 | 4 | |
| Pre-operative self-esteem | 6 | 6 | 6 | 6 | |
| Post-operative 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 |

The numbers are expressed in median. For testing of total score the Friedman test was used for 4 paired samples of semi-continuous variable. Significance was set on p-value < 0.05.

Table 2. Mean scores SSQ over time

| SSQ | 6 weeks (T1) | T1 versus T2 p-value | 6 months (T2) | T2 versus T3 p-value | 1 year (T3) | T3 versus T4 p-value | 2 year (T4) | Overall p-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 pre-operative 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 post-operative 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 |
| Pre-operative 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 |
| Post-operative 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 |

table continues

| SSQ | 6 weeks (T1) | T1 versus T2 p-value | 6 months (T2) | T2 versus T3 p-value | 1 year (T3) | T3 versus T4 p-value | 2 year (T4) | Overall p-value |
|--|-----------------|-------------------------------|---------------------|-------------------------------|-----------------|-------------------------------|-----------------|--------------------|
| 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) | 0.090 | 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 |

The numbers are expressed in means and SD. For testing of scores the Friedman test was used for 4 paired samples of semi-continuous 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 on p-value < 0.05.

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).

Discussion

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 [3,4]. 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 [5]. However, in general, studies measuring quality of life and body image show that these items are not stable over time [6,7]. 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 ones seemed doubtful.

To come to sound conclusions the differences between the study set-up of Krasopoulos 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 [1]. 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 two 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 significant 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 post-operative were at an increased level at 6 weeks compared to preoperative and decreased afterwards.

Pain is also present in the normal non-surgical adolescent school population. A study of Haraldstad et al. showed that pain problems are highly prevalent in adolescents and health related quality of life scores (HRQoL) are impaired in adolescents with pain compared to peers without pain. Girls were more affected than boys [8]. 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, the pain diminished and thus 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 [9].

Furthermore, healthy school children show differences in health-related quality of life over time which seems merely influenced by age and sex [10]. 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 a pectus excavatum responses on serial questionnaires would lead to different scores over time as a reflection of the phase the adolescent is in [11]. This could also have an effect on the outcome measurement 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 [12].

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 between 6 weeks and 6 months. Although the differences are statistically significant, the changes after 6 months postoperatively are small and probably not clinical 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 about 3 years. However, since the patient groups undergoing Nuss bar implantation are in their adolescence, it could also be that in order 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 Single Step Questionnaire scores depending on the time of application post-operative. As long as it is not established that patients do reach a steady state after PE correction we recommend that the SSQ questionnaire should not be used as single step questionnaire but administered at different post-operative time intervals, especially before and after 6 months post-operatively, in order to come to a more exact assessment of the overall satisfaction post-operative.

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