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Health-related quality of life after Nuss procedure for pectus excavatum: a cross-sectional study

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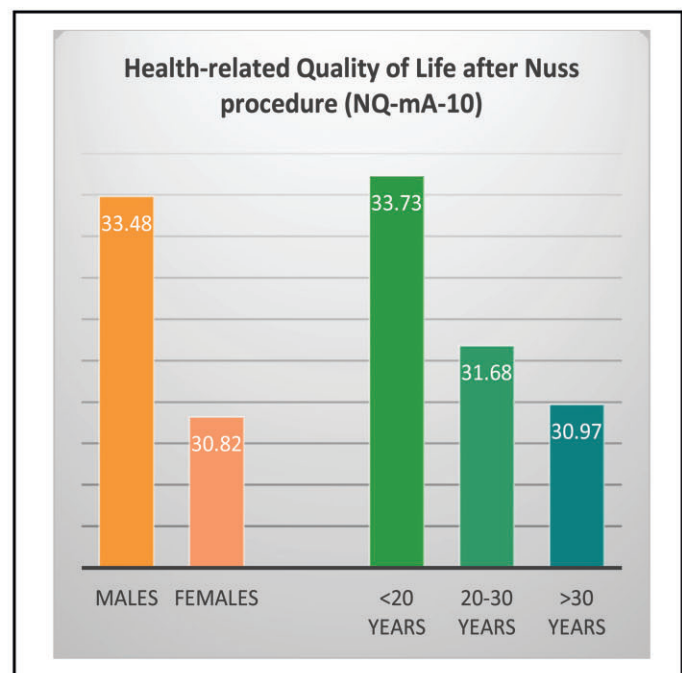
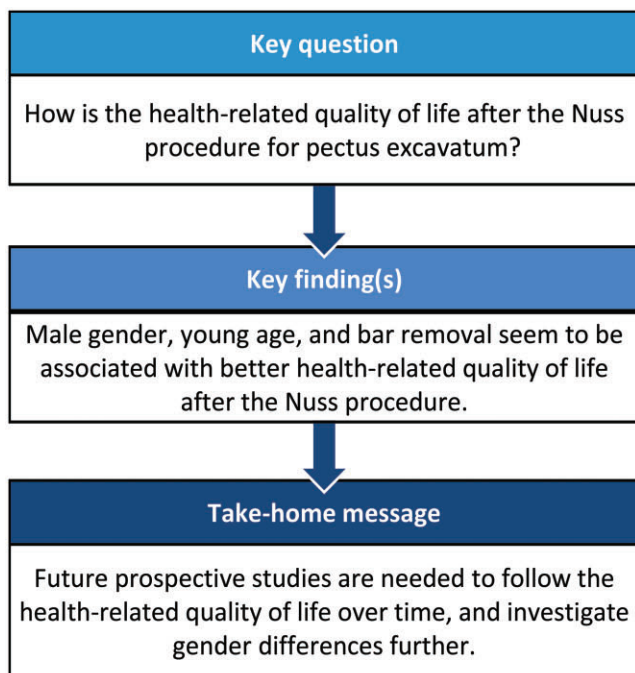
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Abstract

OBJECTIVES: Pectus excavatum (PE) can cause both physical and psychosocial symptoms and affect patients' health-related quality of life. Previous international studies have shown that the Nuss procedure increases both self-esteem and body image. The aim of the study was to evaluate the health-related quality of life in patients who have undergone the Nuss procedure for PE.

METHODS: The study had a cross-sectional multicentre design. All patients ($N = 420$) who underwent the Nuss procedure for PE in 3 cardiothoracic departments in Sweden from 2000 to 2019 were invited to answer the RAND-36 and Nuss Questionnaire modified for adults.

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Genders were compared using the Mann–Whitney *U*-test. Patients were divided into groups based on age at time of surgery (<20, 20–30 or >30 years) and analysed by the Kruskal–Wallis *H*-test with *post-hoc* analyses.

RESULTS: A total of 236 patients returned the questionnaires; 82.2% were males. Men scored significantly better on the modified Nuss Questionnaire total ($P = 0.01$) and psychosocial ($P = 0.02$) subscales. Patients who had surgery at <20 years of age had significantly better scores on the same scales ($P = 0.007$ and 0.006 , respectively) compared to patients aged 20–30 years at the time of surgery. However, no significant difference was seen in comparison with patients >30 years. Patients who had their bar removed had significantly better values on both scales.

CONCLUSIONS: Male gender, young age at surgery and bar removal seem to be associated with better health-related quality of life after the Nuss procedure for PE.

Keywords: Pectus excavatum • Nuss procedure • Quality of life • Bar removal

ABBREVIATIONS

| | |
|-------|--|
| HRQoL | Health-related quality of life |
| NQ-mA | Nuss Questionnaire modified for Adults |
| PE | Pectus excavatum |

INTRODUCTION

Pectus excavatum (PE) represents 90% of all thoracic deformities and occurs in 1 in 400 live births, predominately affecting males [1]. PE is characterized by a depression of the sternum and adjacent costal cartilages [2]. Even though the deformity can sometimes be seen at the time of birth, it often increases as the child grows until culminating and thereafter stabilizing at the time of puberty [3].

PE can cause physical, primarily cardiopulmonary, symptoms, such as dyspnoea, decreased exercise capacity [4] and palpitations [5]. Patients with PE also suffer from psychosocial distress due to the deformity; commonly reported issues are poor body image, low self-esteem [6, 7] and feelings of shame because of how the chest looks [8]. These physical and psychosocial issues cause patients with PE to have decreased health-related quality of life (HRQoL) [8]. Many patients turn to corrective surgery for help. The gold standard surgical technique is a minimally invasive repair known as the Nuss procedure [9]. The procedure involves making 2 lateral incisions bilaterally in the chest wall and placing one or more convex metal bars beneath the sternum under thoroscopic guidance to push the depression outwards [10]. The bar(s) are left in place for 2–4 years [11].

The Nuss procedure was first suggested for a paediatric population, to be performed before puberty due to the more malleable chest wall [10]. The use of the Nuss procedure has also been successful in the adult population despite their more rigid bones [12] and older age being associated with more difficult repairs and increased risk of complications [5, 13, 14]. The Nuss procedure can improve cardiopulmonary function, though these changes seem to have a range of subjective importance [15]. In addition, the procedure has been shown to increase self-esteem and improve body image [16]. Corrective surgery for PE is also associated with improved HRQoL [6, 17].

To achieve a complete overview of the HRQoL in patients with PE, it is advantageous to assess both generic and disease-specific HRQoL because they provide complementary information. The most widely used generic instrument is RAND-36 [18], and the Nuss Questionnaire modified for Adults (NQ-mA) is one of

the most frequently used instruments when assessing disease-specific HRQoL among patients with PE [8, 17, 19, 20].

In Sweden, the number of corrective surgeries for PE has increased. Between 2014 and 2018, ~72 surgeries were performed annually, whereas 124 surgeries were performed in 2019 (https://sdb.socialstyrelsen.se/if_ope/resultat.aspx, 21 April 2021, date last accessed). It is important to evaluate surgical outcomes not only from the perspective of physical results but also the effects on HRQoL. To the best of our knowledge, no studies on this topic have been conducted in a Swedish context, so it would be of interest to evaluate whether the surgical results in Sweden are coherent with international studies. The aim of the present study was to evaluate the HRQoL in patients who have undergone the Nuss procedure for PE and investigate eventual differences among gender and age groups.

PATIENTS AND METHODS

Ethical statement

The study was conducted in accordance with the 1964 Helsinki Declaration and informed consent was obtained from all participants. The study was approved by the Regional Ethical Review Board in Uppsala, Sweden, 19 September 2018 (2018/365, 2018/365/1, 2019-01177, 2019-01177B).

Data collection

This study had a cross-sectional multicentre design. Data were collected between April and May 2019 at the 3 cardiothoracic departments that perform the most Nuss procedures in Sweden. All patients ($N = 420$) included in the study underwent a primary Nuss procedure for PE at Skåne University Hospital in Lund, Sahlgrenska University Hospital in Gothenburg or Örebro University Hospital between 2000 and 2019. Patients who were <15 years old at the start of the study were excluded. Questionnaires were sent out by mail and could be filled out either manually or digitally. A reminder was mailed 2 and 4 weeks after the initial request. The same study sample was previously used to validate the Swedish version of the NQ-mA, which is described in detail elsewhere [21].

Questionnaires

The original NQ-mA is a 12-item disease-specific HRQoL questionnaire for patients with PE [19]. Scores and analyses in the

current study are based on the Swedish validated version of the instrument 'NQ-mA-10' [21], which consists of 10 items divided into 2 subscales: psychosocial and physical. Scores range between 10 and 40 for the overall score on the NQ-mA-10, with a higher score indicating better HRQoL.

The RAND-36 [18] is a 36-item generic HRQoL questionnaire that contains 8 domains: physical functioning, role-physical, bodily pain, general health, vitality, social functioning, role-emotional and mental health. Domain scores range from 0 to 100, with higher scores indicating better HRQoL.

All participants filled out a study-specific form with demographic questions, including sex, age, education, occupation, year of surgery, surgical department, whether their bars had been removed, and, if so, in what year.

Statistical analysis

A non-response analysis was conducted using the multidimensional chi-squared test. Missing data were replaced using person mean substitution [22]. Answering alternatives on both questionnaires were ordinally levelled and non-parametric statistical methods used. Despite this, the data on each subscale are given as percentages or mean \pm standard deviation (SD).

Gender differences were examined using the Mann-Whitney *U*-test. For age comparisons, the participants were divided into 3 different groups based on age at time of surgery: <20, 20–30 and >30 years. For these comparisons, the Kruskal-Wallis *H*-test was applied and *post-hoc* pairwise comparisons performed for all significant values with the Mann-Whitney *U*-test and the Bonferroni correction to adjust for multiple testing. To examine whether any differences were present between those who had or had not yet had their bar(s) removed, the Mann-Whitney *U*-test was applied. Significance was defined as $P < 0.05$. Statistical analyses were performed using IBM SPSS Statistics version 25.0 [23].

RESULTS

Out of the 420 potential participants, 236 returned the questionnaires, giving a response rate of 56.2%. Three of the responders only responded to the RAND-36. These cases were considered to be missing and were not included in the analysis. One patient identified themselves as neither male nor female and was not included in the gender comparisons. Males were significantly more likely to be non-responders [χ^2 (1, $N = 419$) = 5.83, $P = 0.016$], but the measure of association was weak ($\phi = 0.116$). No association was found between non-response and age [χ^2 (2, $N = 420$) = 2.80, $P = 0.246$] or between non-response and years since surgery [χ^2 (3, $N = 420$) = 5.63, $P = 0.131$]. Patient characteristics are presented in Table 1.

Male participants scored significantly higher on the NQ-mA-10 in regards to the total score and psychosocial subscale compared to female participants, but we found no significant difference between genders on the physical subscale or any of the domains of the RAND-36 (Table 2). Differences in the HRQoL scores for different age groups are presented in Table 3. The analysis revealed a significant difference in the NQ-mA-10 total and psychosocial scores, and *post-hoc* tests showed a significant difference between patients aged <20 years and those aged 20–30 years at the time of surgery. We also identified significant differences between the same age groups in the RAND-36

Table 1: Patient characteristics

| N | 236 |
|---|------------------------|
| Sex | |
| Male | 194 (82.2) |
| Female | 41 (17.4) |
| Other | 1 (0.1) |
| Mean age, years \pm SD (min-max) | 25.9 \pm 7.1 (15–67) |
| Mean age at surgery, years \pm SD (min-max) | 19.3 \pm 5.4 (10–57) |
| Education | |
| Compulsory | 50 (21.2) |
| Upper secondary | 116 (49.2) |
| Higher vocational education | 19 (8.1) |
| University | 50 (21.2) |
| Missing | 1 (0.4) |
| Occupation | |
| Employed | 144 (61.0) |
| Student | 80 (33.9) |
| Job applicant | 8 (3.4) |
| Sick leave | 3 (1.3) |
| Missing | 1 (0.4) |
| Year of surgery | |
| 2000–2004 | 21 (8.9) |
| 2005–2009 | 49 (20.8) |
| 2010–2014 | 71 (30.1) |
| 2015–2019 | 95 (40.2) |
| Department | |
| Gothenburg | 62 |
| Lund | 85 |
| Örebro | 89 |
| Bar(s) removed | 152 (64.4) |

Data are given as *n* (%) unless otherwise noted.

domains social functioning and mental health. *Post-hoc* tests revealed that the significance was between those who were aged <20 years and those aged 20–30 years at the time of surgery. No significant differences were found for patients who were aged >30 years compared either of the other age groups.

Participants who had not yet had their bar(s) removed comprised 35.6% of the total study population. These patients scored significantly lower on the NQ-mA-10 physical subscale and the RAND-36 domains physical functioning, role-physical and bodily pain compared to patients who had had their bar(s) removed (Table 4).

DISCUSSION

This study evaluated the HRQoL in patients who had undergone the Nuss procedure for PE in Sweden and investigated eventual differences among gender and age groups. This is the first study to use NQ-mA-10 for analysis since its validation [21]. The main findings of our study were that women and patients having the surgery between 20 and 30 years of age have significantly lower total and psychosocial disease-specific HRQoL compared to men and those having the surgery at <20 years of age, respectively. In addition, patients who still have their bar(s) in place have lower physical HRQoL, both generic and disease-specific.

Our finding that women scored lower NQ-mA-10 psychosocial and total scores than men is a new addition to previous international findings. Jacobsen *et al.* [20] and Metzelder *et al.* [24] found no gender differences when assessing generic and disease-specific HRQoL. Reference data state that men have better generic HRQoL [25], however, this is contrary to our results as it

Table 2: Total and subscale scores for NQ-mA-10 and RAND-36 with a comparison between genders

| | Mean ± SD | Min-max | Skewness | Kurtosis | Male (n = 191) Mean ± SD | Female (n = 41) Mean ± SD | U | P-value |
|-----------------|-------------|---------|----------|----------|--------------------------------|---------------------------------|---------|---------|
| NQ-mA-10 | | | | | | | | |
| Total | 33.0 ± 5.88 | 14–40 | -1.12 | 0.80 | 33.48 ± 5.61 | 30.82 ± 6.63 | 2811.00 | 0.01 |
| Psychosocial | 23.1 ± 4.74 | 8–28 | -1.23 | 0.90 | 23.46 ± 4.50 | 21.46 ± 5.53 | 2988.50 | 0.02 |
| Physical | 9.9 ± 9.00 | 3–12 | -1.14 | 0.87 | 10.02 ± 1.94 | 9.37 ± 2.33 | 3338.50 | 0.10 |
| RAND-36 | | | | | | | | |
| PF | 92.7 ± 14.6 | 5–100 | -3.26 | 12.13 | 92.7 ± 15.04 | 91.9 ± 13.00 | 3599.50 | 0.40 |
| RP | 85.2 ± 29.6 | 0–100 | -1.90 | 2.32 | 85.1 ± 29.31 | 84.4 ± 31.87 | 3931.50 | 0.99 |
| BP | 85.3 ± 20.6 | 10–100 | -1.53 | 1.76 | 85.8 ± 20.49 | 82.5 ± 21.62 | 3467.50 | 0.30 |
| GH | 72.9 ± 20.0 | 10–100 | -0.74 | 0.27 | 74.3 ± 18.77 | 68.4 ± 23.77 | 3424.50 | 0.30 |
| VT | 60.6 ± 18.6 | 5–100 | -0.46 | 0.08 | 61.2 ± 18.57 | 58.0 ± 18.50 | 3451.00 | 0.30 |
| SF | 83.9 ± 22.1 | 0–100 | -1.53 | 2.00 | 83.9 ± 21.95 | 83.1 ± 23.78 | 3723.50 | 0.70 |
| RE | 79.0 ± 32.8 | 0–100 | -1.32 | 0.38 | 79.3 ± 32.41 | 77.5 ± 34.91 | 3887.00 | 0.90 |
| MH | 74.3 ± 17.2 | 8–100 | -0.94 | 0.88 | 74.7 ± 17.28 | 72.4 ± 17.47 | 2490.00 | 0.40 |

BP: bodily pain; GH: general health; MH: mental health; NQ-mA-10: Nuss questionnaire modified for adults 10-item version; PF: physical functioning; RE: role-emotional; RP: role-physical; SF: social functioning; VT: vitality.

Table 3: Health-related quality of life differences based on age at surgery

| | Age | | | H (df) | P-value |
|-----------------|----------------------------|----------------------------|-----------------------|-----------|---------|
| | <20 years (n = 156) | 20–30 years (n = 68) | >30 years (n = 12) | | |
| NQ-mA-10 | | | | | |
| Total | 33.73 ± 5.79 ^a | 31.68 ± 5.60 ^a | 30.97 ± 6.96 | 10.06 (2) | 0.007 |
| Psychosocial | 23.69 ± 4.60 ^b | 22.10 ± 4.68 ^b | 21.22 ± 5.31 | 10.23 (2) | 0.006 |
| Physical | 10.05 ± 1.70 | 9.58 ± 2.12 | 9.75 ± 2.67 | 2.87 (2) | 0.20 |
| RAND-36 | | | | | |
| PF | 93.05 ± 14.62 | 92.50 ± 13.85 | 87.08 ± 20.94 | 1.79 (2) | 0.40 |
| RP | 87.25 ± 26.57 | 81.82 ± 33.84 | 81.25 ± 35.56 | 0.48 (2) | 0.80 |
| BP | 86.72 ± 19.26 | 82.95 ± 22.80 | 87.50 ± 19.63 | 1.68 (2) | 0.40 |
| GH | 75.13 ± 20.06 | 70.02 ± 18.74 | 70.42 ± 18.76 | 3.65 (2) | 0.20 |
| VT | 62.83 ± 18.4 | 56.67 ± 17.48 | 59.17 ± 20.65 | 5.44 (2) | 0.06 |
| SF | 86.41 ± 20.49 ^c | 78.22 ± 25.13 ^c | 84.38 ± 22.69 | 7.83 (2) | 0.02 |
| RE | 80.76 ± 32.46 | 73.23 ± 35.19 | 86.11 ± 22.29 | 3.81 (2) | 0.20 |
| MH | 76.21 ± 16.83 ^d | 70.06 ± 18.46 ^d | 76.00 ± 13.54 | 6.24 (2) | 0.04 |

Data are given as mean ± SD. Significant pairwise comparisons are marked with the same superscript letter.

BP: bodily pain; GH: general health; MH: mental health; NQ-mA-10: Nuss questionnaire modified for adults 10-item version; PF: physical functioning; RE: role-emotional; RP: role-physical; SF: social functioning; VT: vitality.

was only the disease-specific HRQoL that differed. A possible explanation of women scoring lower on the total and psychosocial scale could be associated to women worrying more and reporting higher levels of depression due to visible disfigurement than men [26], which may tend to have effects even after surgery. Future studies with a qualitative focus would potentially be useful in further investigating gender differences.

The optimal age for performing the Nuss procedure has been unclear [9]. We found that patients who underwent the Nuss procedure at <20 years old had better HRQoL than those who underwent the procedure between the age of 20 and 30 years. This difference was also visible in the RAND-36 domains social functioning and mental health. Though the difference was not significant, those undergoing the Nuss procedure after the age of 30 years had a tendency to have lower NQ-mA-10 total and psychosocial subscale scores compared to the other groups. The less favourable outcome in the >30 years age group is in line with

previous reports of higher age being associated with more difficult repairs and increased risk of complications [5, 13, 14]. The lack of significant differences in our study may be due to the few patients in this group. Despite this, we chose to maintain our group divisions as previous research [5] and our own clinical experience, emphasizes the more complicated repairs patients >30 years faces.

Our results also demonstrate that patients with their bars still in place have significantly lower scores on the physical subscales, both the generic and disease-specific assessments. Our results could potentially correlate with the pain many patients experience because of the bars. Ninety-five per cent of all patients who have undergone the Nuss procedure for PE report some level of pain caused by the bars, whereas 85% state that there is no pain after bar removal [14]. Yet, our results are not in agreement with previous findings. Kim *et al.* [27] investigated HRQoL before the Nuss procedure, after surgery, and after bar removal and found

Table 4: Health-related quality of life differences in patients with or without bar removal

| | Removed (n = 152) | Not removed (n = 84) | U | P- value |
|-----------------|----------------------|-------------------------|---------|-------------|
| NQ-mA-10 | | | | |
| Total | 33.17 ± 5.94 | 32.66 ± 5.75 | 5885.50 | 0.50 |
| Psychosocial | 22.87 ± 4.87 | 23.52 ± 4.41 | 6835.00 | 0.20 |
| Physical | 10.29 ± 1.83 | 9.14 ± 2.25 | 4181.50 | <0.001 |
| RAND-36 | | | | |
| PF | 95.16 ± 13.08 | 87.63 ± 16.55 | 3294.00 | <0.001 |
| RP | 91.95 ± 23.66 | 72.76 ± 34.70 | 4301.50 | <0.001 |
| BP | 90.52 ± 16.87 | 76.41 ± 23.15 | 3637.50 | <0.001 |
| GH | 73.28 ± 19.86 | 73.61 ± 19.46 | 6197.00 | 0.90 |
| VT | 60.83 ± 17.87 | 60.85 ± 19.46 | 6099.50 | 0.90 |
| SF | 83.89 ± 23.53 | 83.97 ± 19.73 | 5986.00 | 0.50 |
| RE | 78.75 ± 33.61 | 79.10 ± 31.83 | 6152.00 | 0.90 |
| MH | 71.80 ± 17.30 | 74.05 ± 17.47 | 5938.50 | 0.60 |

Data are given as mean ± SD.

BP: bodily pain; GH: general health; MH: mental health; NQ-mA-10: Nuss questionnaire modified for adults 10-item version; PF: physical functioning; RE: role-emotional; RP: role-physical; SF: social functioning; VT: vitality.

no improvements in HRQoL after bar removal. However, they only assessed items regarding pain after bar removal. Therefore, it is possible that the items assessed at all time-points were not able to catch the aspects of pain that correlate with the bar and could potentially explain the difference from our results.

The major strength of this study is the large study sample, which allows for greater knowledge of how generic and disease-specific HRQoL is perceived in patients who have undergone the Nuss procedure for PE in a Swedish setting.

Limitations

One limitation due to the cross-sectional design is that comparisons of HRQoL over time were not possible. However, this design was considered the most suitable for describing the overall HRQoL of the majority of patients who underwent the Nuss procedure for PE in Sweden. Further limitations are the response rate and the unknown characteristics of the non-responders and eventual non-response bias. Previous international studies have reported different response rates of 54% [28] and 70% [29]. The latter study administered questionnaires by telephone, which may have positively affected the response rate. However, response rate has been discussed to have little association with the eventual non-response bias [30]. The 20-year time range for when the patients underwent surgery could be a potential cause of recall bias, but both questionnaires ask for current health status and should not be affected by failed memory. Yet, the results of this study have raised questions for future research: it is essential to follow these patients over time, for HRQoL assessments pre- and post-surgery, as well as after bar removal. Further investigation of differences in assessment between men and women are also of interest.

Conclusion

In conclusion, this study shows that the generic and disease-specific HRQoL in patients who have undergone the Nuss

procedure for PE in Sweden differs in different subgroups. Male gender, young age and bar removal seem to be associated with better HRQoL. Further studies are required to observe longitudinal data and confirm our findings.

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Author contributions

Louise Norlander: Conceptualization; Data curation; Formal analysis; Methodology; Writing—original draft; Writing—review & editing. **Ann-Sofie Sundqvist:** Conceptualization; Project administration; Supervision; Writing—review & editing. **Agneta Anderzén-Carlsson:** Conceptualization; Supervision; Writing—review & editing. **Mats Dreifaldt:** Conceptualization; Writing—review & editing. **Jesper Andreasson:** Conceptualization; Writing—review & editing. **Mårten Vidlund:** Formal analysis; Methodology; Supervision; Writing—review & editing.

Reviewer information

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