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## Seminars in Pediatric Surgery

journal homepage: [www.elsevier.com/locate/sempedsurg](http://www.elsevier.com/locate/sempedsurg)

# Outcomes, quality of life, and long-term results after pectus repair from around the globe

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## ARTICLE INFO

### Keywords:

Pectus excavatum  
Pectus carinatum  
Nuss procedure  
Ravitch operation  
Body image  
Quality of life  
Outcomes  
Long-term results

## ABSTRACT

The procedure introduced by Donald Nuss in 1997 at the American Pediatric Surgical Association meeting has now been adopted by pediatric, thoracic, and plastic surgeons around the globe. Since its introduction, the Nuss procedure has been the subject of intense scrutiny. More than 20 years since the original publication, medical centers from around the world have reported their experience with the procedure and robust evidence now supports its safety and efficacy.

Additionally, in collaboration with psychologists in Norfolk, a methodology was devised to measure the psychosocial as well as the physical benefits of the procedure. Extensive independent multinational and multi-institutional data demonstrate repair of pectus excavatum using the Nuss procedure results in marked improvement in both physical function and body image. This report reviews the outcomes, quality of life, and long-term results of numerous centers worldwide since the introduction of the procedure.

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## Introduction

From the very introduction of his procedure, Donald Nuss and his colleagues at Children's Hospital of The King's Daughters in Norfolk, Virginia, USA have been concerned about evaluating and objectively reporting the outcomes and long-term results of the operation. In the 1998 report, Nuss waited to introduce the technique until 10 years of experience had been gained with it, and 30 of 42 initial patients had already undergone bar removal by the time of the initial publication.<sup>1</sup> In that report, "Initial excellent results were maintained in 22, good results in four, fair in two, and poor in two, with mean follow-up since surgery of 4.6 years." Subsequently, we have reported our results periodically in the literature.<sup>2,3</sup> In the 2010 report, "Twenty-One Years of Experience With Minimally Invasive Repair of Pectus Excavatum by the Nuss Procedure in 1215 Patients", a good or excellent anatomic surgical outcome was achieved in 95.8% of patients at the time of bar removal.<sup>3</sup> A fair result occurred in 1.4%, poor in 0.8%, and recurrence of sufficient severity to require reoperation occurred in 11 (1.4%). Though our reports have always stated our complications, that topic will be discussed in full detail by other authors in this *Seminars in Pediatric Surgery* issue (see Goretsky and McGuire,

pages xx–yy). Here we will review the outcome results obtained by other authors, and discuss the body image and quality of life reports which pertain to the procedure. Since many reports admix anatomic and functional outcomes with body image studies, we will report authors' data on both outcomes, chronologically.

## Methods and results

Early in our experience in Norfolk we were struck by the embarrassment patients felt because of Pectus excavatum. Boys were reluctant to remove their shirts for exam, and frequently families offered spontaneously the observation that the boy would not go swimming in public. It was remarkable, we thought, to see the same patient after surgical correction wearing no shirt in his hospital bed. We noted how proud patients seemed to be at postop visits in the clinic – they would remove their shirt immediately, "like Superman in a phone booth." To learn how to investigate this behavior, we sought a leading authority on body image, Dr. Thomas F. Cash of Old Dominion University, to develop a way to measure this phenomenon. The disease-specific quality of life Pectus Excavatum Evaluation Questionnaire (PEEQ) was developed with Dr. Cash. It was validated with interviews of 19 patients and 22 parents, and was found to have high test-retest reliability. This pilot study showed significant improvements in exercise intolerance, shortness of breath, and tiredness, and decreases in the frequency of the child being frustrated, sad, self-conscious, and

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isolated following surgical correction.<sup>4</sup> Encouraged by these findings, we then utilized this tool as part of a multi-center study of Pectus excavatum conducted together with 11 leading North American pediatric surgical centers, reporting on 247 patients and 274 parents who completed both pre- and post-operative interviews. In that study, preoperative psychosocial functioning was found to be unrelated to objective Pectus excavatum severity (as measured by computed tomographic Haller index). Patients and their parents reported significant postoperative changes. Improvements occurred in both physical and psychosocial function.<sup>5</sup> This psychologic tool was subsequently made available to all without cost through the website of the American Psychological Association PsychTESTS database (website: [www.apa.org](http://www.apa.org)). In addition, we freely gave it to other investigators who requested it from a variety of countries.

Krasopoulos and colleagues, from London, UK, modified that PEEQ for adults. They found in 20 young male adults (14–37 years, median 18 years), after modifying the questionnaire and doing test retest validation on 5 patients, that the domains for social functioning and self-esteem showed highly significant improvement following surgery. They did not find improvement in physical function.<sup>6</sup>

In 2007, Petersen et al., from Hannover, Germany, reported on 40 patients utilizing the methodology developed by Krasopoulos et al for adults.<sup>7</sup> In patients aged 10–24 years, mean age 17 years, they found a high level of persistent satisfaction with Minimally Invasive Repair of Pectus Excavatum (MIRPE) after bar removal, and a highly significant correlation between self and external assessment. Analysis of specific and total scores revealed a significant improvement of psychosocial and physical well-being after bar implantation, which persisted up to 4 years after bar removal. Age and sex had no significant impact on scores in either patients or parents.

In 2008, Skarsgard et al., from Vancouver, British Columbia, Canada, reported a 3-year experience of 43 patients who underwent surgery for Pectus excavatum, and were evaluated for health related quality of life (HRQL) by the Child Health Questionnaire and the PEEQ. Their report compared results after Ravitch and Nuss procedures, and found that patients undergoing surgery for PE by either Nuss or Ravitch procedure had similar clinical and HRQL outcomes, but as a group had poorer HRQL scores than age-matched population norms.<sup>8</sup>

In 2009, Coelho et al. reported on 40 patients with Pectus excavatum from Curitiba, Brazil.<sup>9</sup> With an average follow up of 28 months, they found no significant difference in complications. Among 20 Nuss procedure patients, they experienced “Poor” results in 3, or 15%, which consisted of overcorrection to carinatum. Otherwise, all patients had either “Good” or “Fair” results. Interestingly, in the conclusion the authors report, “In this series, no significant difference was found between sternochondroplasty and Nuss with regards to aesthetic results.” But they favor sternochondroplasty for asymmetric pectus excavatum because of their experience with reactive pectus carinatum. No data were offered regarding recurrence of pectus excavatum in either group.

In 2011, Shu et al., from Hangzhou, China reported experience with 406 patients with Pectus excavatum.<sup>10</sup> All patients were pediatric, with an average age of 7 years. They reported excellent results in 95%, good results in 3%, and fair results in 2%. Follow up was from 3 months to 6 years, and 40% had undergone bar removal. Five patients required redo operation because of scoliosis caused by pain in two, and bar displacement in 3, with resolution in all five.

In 2011, Steinmann et al, from Erlangen, Germany, reported on 90 patients, aged 14–35 years, 71 with Pectus excavatum, and 19 with Pectus carinatum.<sup>11</sup> Patients were first screened by a stan-

dardized Short Diagnostic Interview for Mental Disorders. Neither PE nor PC was associated with increased rates of mental disorders. In 14% of PE patients and 5% of PC patients, full criteria for a current diagnosis of mental disorder were found. Subsequently, patients were evaluated with a Short-Form 36 Health Survey, the Nuss Questionnaire Modified for Adults, a separate Body Image Questionnaire, and finally a Dysmorphic Concern Questionnaire. Afterward, self-assessment of physical appearance, self-esteem, and depression were obtained. Notably, before operation, “almost every PE patient reported physical symptoms...only 3.2% of PE patients displayed no symptoms.” Patients were asked for internal and external motivational factors to undergo surgery. A combination of both cosmetic and physical health-related reasons was cited in 51%; in 11%, cosmetic reasons alone; and physical health alone in 24%. Unfortunately, this very thorough study did not assess patients after operation.

Also in 2011, Kim et al., from Seoul, Korea reported on 39 patients, mean age 7 years old, before and after the Nuss procedure, and after bar removal.<sup>12</sup> They were studied by a questionnaire which evaluated satisfaction with chest appearance, social belonging, self-confidence, physical stamina, and other variables. Marked improvements in all variables were seen between preoperative and post Nuss procedure, with high internal consistency. However, there were no further improvements after bar removal, suggesting that it is the repositioning of the sternum which led to the improvements.

In 2011, Hollwarth et al., from Graz, Austria, reported on 17 patients tested in 2003 and again in 2007.<sup>13</sup> They used the Operation-Expectation-Questionnaire, developed for the study; the Frankfurter Body concept Scales, and the Symptom-Checklist Revised. They concluded that “The long-time follow-up can make us sure, that the Nuss procedure as a physical treatment has positive effects on physical as well as psychological aspects of young adults. The patients’ confidence with the cosmetic result was very high, which reflects the excellent effects of the minimal invasive repair according to Nuss.”

In 2013, Yuksel and colleagues, from Istanbul, Turkey, reported on 30 patients, median age 14 years, who had undergone minimally invasive repair of Pectus carinatum.<sup>14</sup> They utilized a modified Nuss questionnaire, both for patients and parents, translated into Turkish. There was a marked improvement in both psychosocial and physical scores after surgical correction.

In 2014, the same Istanbul group reported on quality of life in Pectus excavatum, describing 88 patients, mean age 18 years.<sup>15</sup> They again utilized a modification of the Krasopoulos questionnaire, translated into Turkish. Psychosocial change was notably greater in the patients’ than the parents’ assessment. However, the physical changes and Nuss questionnaire changes confirmed previous reports.

In 2014, Colombani and colleagues, from Baltimore, Maryland, USA, reported results of 336 patients treated by Nuss procedure.<sup>16</sup> They modified the Nuss procedure by omitting thoracoscopy, doing a retrosternal dissection via a left-to-right thoracic approach, and utilized four-point stabilization of the bar. They reported no cardiac perforations, and pericarditis in 0.6%. Bar displacement had fallen to 1.2%. Recurrence occurred in 2 patients (0.6%), who required reoperation.

Also in 2014, Jouve et al., from Marseille, France, reported on outcomes of the Nuss procedure in 70 patients with average age 14 years.<sup>17</sup> They reported that the cosmetic outcome was considered satisfactory by the patients in 91% of cases, and by the surgeon in 86%. They state, “The minimal scarring and reliably good outcomes support the widespread use of the Nuss technique in children and adolescents.”

In 2015, Colombani, Abdullah, and colleagues reported on 264 patients who underwent repair of Pectus excavatum in 2012 and

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