Advancing pectus deformity care: evaluation of current treatments, complications and future innovations

Braak, H. van

ADVANCING PECTUS DEFORMITY CARE

EVALUATION OF CURRENT TREATMENTS, COMPLICATIONS AND FUTURE INNOVATIONS

Hendrik van Braak

ADVANCING PECTUS DEFORMITY CARE:
EVALUATION OF CURRENT TREATMENTS, COMPLICATIONS AND FUTURE INNOVATIONS

Hendrik van Braak

Advancing Pectus Deformity Care: Evaluation of Current Treatments, Complications and Future Innovations
Thesis, Leiden University, the Netherlands

Provided by thesis specialist Ridderprint, ridderprint.nl
Printing: Ridderprint
Cover and chapter spread design: Heiltje van Braak - van Manen
Layout: Froukje Frijlink, persoonlijkproefschrift.nl

ISBN: 978-94-6506-583-0

Advancing Pectus Deformity Care: Evaluation of Current Treatments, Complications and Future Innovations

Proefschrift

ter verkrijging van
de graad van doctor aan de Universiteit Leiden,
op gezag van rector magnificus prof. dr. S. de Rijcke,
volgens besluit van het college voor promoties
te verdedigen op woensdag 11 maart 2026
klokke 10:00 uur.
door
Hendrik van Braak
geboren te Veenendaal
in 1997

Promotiecommissie

Promotor

Prof. dr. L.W.E. van Heurn

Copromotor

Dr. J.R. de Jong AMC-UvA

Leden promotiecommissie

Prof. dr. N.D. Bouvy

Prof. dr. F.M. Haecker University of Basel

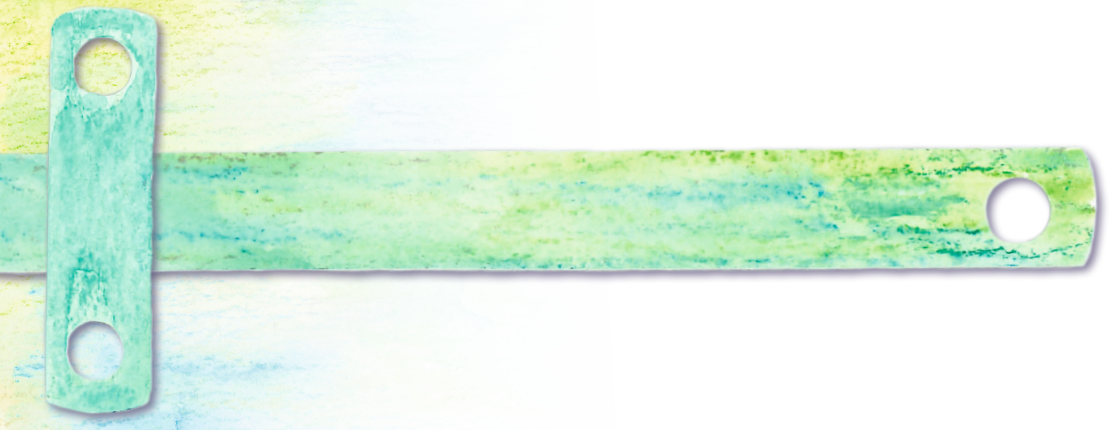
Prof. dr. J.P.M. Derikx

Prof. dr. I.B. Schipper

Dr. E.R. de Loos Maastricht University

Financial support by **Pampamed, Department of Pediatric Surgery of the Amsterdam University Medical Center, AtriCure, Metrum Cryoflex** and **ChipSoft** for the printing of this thesis is gratefully acknowledged.

CONTENTS



Chapter 1

General introduction and thesis outline

GENERAL INTRODUCTION

This thesis is about pectus deformities. Such chest wall conditions were recognized as early as antiquity, with reports on pectus excavatum dating back to the sixteenth century.[1] Pectus deformities are typically classified into three types: pectus carinatum (PC), or pigeon breast; pectus excavatum (PE), also known as funnel chest; and pectus arcuatum (PA). Each type requires distinct treatment methods.

Pectus carinatum

PC is characterized by the overgrowth of costal cartilage resulting in anterior protrusion of the sternum. It has an incidence ranging from 0.04% to 0.7% in the current literature, with a higher prevalence in males.[2] Although patients are typically asymptomatic, PC is strongly associated with body image disturbance and reduced quality of life.[3-5] Therefore treatment is primarily initiated to enhance body image and quality of life, and for cosmetic reasons.

Treatment

Initially, PC was treated using a traditional, invasive surgical approach; the Ravitch procedure which was first introduced in 1960 and has been modified over the years.[6, 7] Later on, the minimally invasive Abramson procedure was introduced, which is a 'reverse' Nuss procedure. After introduction, this treatment method gradually became the surgical treatment of first choice.[8]

Despite the effectiveness of above surgical options, physicians began to explore non-invasive alternatives early on. Already from the seventies onwards, orthotic treatment using a brace was introduced as an alternative for surgery, with success rates ranging from 40-90%. In addition to the varying success rate, bracing was often complicated by long treatment duration and complications such as discomfort, pain, and skin related problems.[9-13]

In 2008, the Dynamic Compression System brace (DCS-bracing) was introduced.[14] Unlike previous braces, initial research showed DCS-bracing to be highly effective, with reported success rates of up to 90%, with minimal complications, leading to its rapid adoption.[15]

Current challenges

Despite these promising initial outcomes, researchers have not yet compared the results of invasive surgical correction with non-invasive DCS-bracing. Moreover, data on the impact of DCS-bracing on quality of life is limited, with only a few studies addressing this aspect. [12, 16, 17] However, these studies used different types of braces, making comparisons challenging. The limited research on the impact of bracing on quality of life is surprising, given that this is the primary reason patients seek treatment.

Furthermore, it is difficult to determine objective outcomes after bracing since PC is mainly a cosmetic deformity, in contrast to PE. Therefore outcomes ultimately hinge on the patients' own view of success and satisfaction.[18, 19] Clinicians and patients are likely to have differing opinions on what constitutes a successful correction of PC. Differences in such treatment success expectations between patients and physicians can influence treatment decisions. Yet, to date those differences have not been investigated.

Using objective measurements like 3D scanning for monitoring PC offers significant advantages in both clinical decision-making and patient compliance. By providing clear, measurable data on chest wall deformity through the (external) Haller Index ((E)HI),[9, 20, 21] 3D scanning enables clinicians to track treatment progress without radiation exposure. Additionally, having objective data helps patients see their own progress, which can motivate adherence to non-surgical treatments like bracing. However, for these measurements to be fully effective, there is a critical need for reference values that define normal and abnormal (E)HI ranges across age groups and genders. Reference values allow clinicians to set realistic treatment goals and provide benchmarks for success, making it easier to evaluate the impact of interventions and improving consistency in care. These reference values can also be valuable for assessing treatment outcomes in vacuum bell (VB) therapy for pectus excavatum (PE).

Pectus excavatum

PE is characterized by a sunken sternum due to rib cartilage overgrowth. It has an estimated incidence of 0.1-0.8%.[22] While most studies indicate that PE is more common in males, recent literature suggests it may actually be more prevalent in females.[23] However, many females do not seek medical attention, leading to substantial underreporting in this group. Many adult patients experience physical symptoms such as exercise intolerance, chest pain, and respiratory difficulties, though these symptoms often do not appear until puberty.[23, 24] Patients often realize only after treatment that their exercise tolerance had actually been lower beforehand and improved after treatment, though they were not fully aware of the limitation initially. Most pediatric patients with PE are asymptomatic. Primary reasons for pediatric patients to seek

treatment are psychological distress, body image concerns, and reduced quality of life.[4, 25]

Treatment

Currently, minimally invasive repair of PE (MIRPE), also known as the Nuss procedure, is the standard surgical treatment for PE. MIRPE offers excellent long-term outcomes. Numerous techniques and materials are available for the Nuss procedure, leading to considerable variation in practice.[26] In our clinic in Amsterdam, we are using the short bar technique as described by Pilegaard.[27]

An alternative, non-surgical treatment option is VB therapy, which is usually used in patients with relatively mild deformities.[28]

Current challenges

While the surgical technique for the Nuss procedure is well established, postoperative pain management remains a challenge. Pain management following the Nuss procedure is traditionally handled with continuous epidural analgesia (CEA), but intercostal nerve cryoablation has emerged as a promising alternative, potentially reducing opioid use and hospital stay duration.[29] Despite promising results of intercostal nerve cryoablation, long-term data on complications like neuropathic pain remain sparse and other complications potentially related to cryotherapy, such as pleural effusion and hyperinflammatory reactions following cryotherapy, remain underdiscussed in the literature.[30-32]

In addition to the challenges with pain management, complications such as dislocations and infections remain a concern. Dislocations can be reduced by adapting the surgical technique. In case of an postoperative infection, the Nuss bar remains in situ in most instances, but affected patients often require extended antibiotic therapy and, occasionally, additional surgical interventions. Furthermore, material allergies related to the Nuss bar can sometimes be misdiagnosed as infections, leading to complicated clinical courses. The current literature shows no clear consensus on the choice of antibiotics or the optimal duration of treatment.[33-36]

Pectus arcuatum

Pectus arcuatum (PA) is a distinct condition that combines features of both PC and PE, but differs fundamentally in origin. Unlike PE and PC, which are caused by rib cartilage overgrowth, PA results from the premature fusion of the sternal sutures. This leads to an anterior protrusion of the upper sternum, while the lower sternum is typically neutral or indented. Additionally, the sternum in PA is abnormally short.[37]

Treatment

A Ravitch procedure is necessary to correct this complex deformity, PA cannot be corrected with non-invasive treatment.[37]

Cross-cutting perspectives

An additional challenge in the management of pectus deformities lies in determining the optimal timing of treatment. While early intervention is often recommended to take advantage of chest wall flexibility in adolescence, questions remain about how age and psychological development influence patients' motivation to undergo surgery. During periods of prolonged surgical waiting times, particularly during the COVID-19 pandemic, it was observed that a notable number of patients who reached adulthood while awaiting treatment ultimately declined surgery. These observations raise questions about the potential influence of waiting time, increasing cognitive maturity, evolving self-image, and shifting personal priorities on patients' treatment decisions. Despite its clinical relevance, this topic has not yet been studied.

Aims

The aims of this thesis are to present the long-term outcomes of treatment for both PE and PC, establish a foundation for further innovations in bracing therapy, provide guidance for managing postoperative complications following the Nuss procedure and explore factors influencing treatment decision-making in the context of prolonged waiting times.

THESIS OUTLINE

Part I explores treatment options and long-terms results for PC. **Chapter 2** compares results of DCS-bracing to Ravitch surgery, advocating DCS-bracing as the preferred first-line treatment. **Chapter 3** examines the impact of DCS-bracing on quality of life in patients with PC, demonstrating a positive effect on patients' quality of life. **Chapter 4** addresses the challenge of assessing treatment outcomes for DCS-bracing by evaluating intra- and interobserver agreement on treatment outcomes and pre-treatment severity, highlighting the need for objective outcomes measures. **Chapter 5** builds on these recommendations, investigating the use of 3D scanning to establish reference values for a normal chest wall by comparing 3D scans of healthy children with those of patients with PC. These reference values offer an objective benchmark for assessing treatment success.

Part II examines treatment options for PE and addresses pain management and the management of complications associated with the Nuss procedure. **Chapter 6** outlines the long-term outcomes of VB therapy, highlighting that longer treatment duration and extended daily use are key factors for success. It demonstrates that older patients with a stiffer chest wall can also achieve positive results if highly motivated and compliant. The chapter also shows that VB therapy remains challenging in women. **Chapter 7** compares intercostal nerve cryoablation with continuous epidural analgesia, demonstrating it as a superior analgesic method. **Chapter 8** focuses on hyperinflammatory pleural response resembling complex pneumonia following the Nuss procedure, proposing a guideline for diagnosis and treatment. **Chapter 9** provides an overview of postoperative infections and allergies following the Nuss procedure and presents a novel guideline for their diagnosis and management.

Part III presents **Chapter 10**, which reports an investigation of the impact of waiting times on pediatric patients' decisions to decline pectus surgery. It shows that over half of patients withdrew after prolonged waiting, with younger age and absence of physical symptoms at waitlist entry as predictors. Main reasons included increased body acceptance and physical development through strength training or weight gain, underscoring the need for reconsideration of current treatment strategies, including the integration of psychological counseling.

Part IV provides a summary of the thesis and discusses future directions and advancements in the field of pectus deformities.

REFERENCES

1 Bauhinus J. Observatorium medicarum, rararum, novarum, admirabilium, et montrosarum, liber secundus. Partibus Vitalibus Thorace Contentis Frankf.1609.

2. Janssen N, Coorens NA, Franssen A, et al. Pectus excavatum and carinatum: a narrative review of epidemiology, etiopathogenesis, clinical features, and classification. J Thorac Dis. 2024;16(2):1687-701.

3. Krille S, Müller A, Steinmann C, et al. Self- and social perception of physical appearance in chest wall deformity. Body Image. 2012;9(2):246-52.

4. Steinmann C, Krille S, Mueller A, et al. 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. Eur J Cardiothorac Surg. 2011;40(5):1138-45.

5. Fortmann C, Petersen C. Surgery for Deformities of the Thoracic Wall: No More than Strengthening the Patient's Self-Esteem? Eur J Pediatr Surg. 2018;28(4):355-60.

6. Ravitch MM. Operative Correction of Pectus Carinatum (Pigeon Breast). Ann Surg. 1960;151(5):705-14.

7. Welch KJ, Vos A. Surgical correction of pectus carinatum (pigeon breast). J Pediatr Surg. 1973;8(5):659-67.

8. Geraedts TCM, Daemen JHT, Vissers YLJ, et al. Minimally invasive repair of pectus carinatum by the Abramson method: A systematic review. J Pediatr Surg. 2022;57(10):325-32.

9. Wong KE, Gorton GE, 3rd, Tashjian DB, et al. Evaluation of the treatment of pectus carinatum with compressive orthotic bracing using three dimensional body scans. J Pediatr Surg. 2014;49(6):924-7.

10. Loff S, Sauter H, Wirth T, Otte R. Highly Efficient Conservative Treatment of Pectus Carinatum in Compliant Patients. Eur J Pediatr Surg. 2015;25(5):421-4.

11. Lee RT, Moorman S, Schneider M, Sigalet DL. Bracing is an effective therapy for pectus carinatum: interim results. J Pediatr Surg. 2013;48(1):184-90.

12. Colozza S, Bütter A. Bracing in pediatric patients with pectus carinatum is effective and improves quality of life. J Pediatr Surg. 2013;48(5):1055-9.

13. Frey AS, Garcia VF, Brown RL, et al. Nonoperative management of pectus carinatum. J Pediatr Surg. 2006;41(1):40-5.

14. Martinez-Ferro M, Bellia Munzon G, Fraire C, et al. Non-surgical treatment of pectus carinatum with the FMF(®) Dynamic Compressor System. J Vis Surg. 2016;2:57.

15. Martinez-Ferro M, Fraire C, Bernard S. Dynamic compression system for the correction of pectus carinatum. Semin Pediatr Surg. 2008;17(3):194-200.

16. Ermerak NO. Compressive external bracing in pectus carinatum : Results of the first 100 patients. Marmara Med J. 2023;36(3):334-8.

17. Fraser S, Harling L, Patel A, et al. External Compressive Bracing With Initial Reduction of Pectus Carinatum: Compliance Is the Key. Ann Thorac Surg. 2020;109(2):413-9.

18. De Beer SA, Blom YE, Lopez M, de Jong JR. Measured dynamic compression for pectus carinatum: A systematic review. Semin Pediatr Surg. 2018;27(3):175-82.

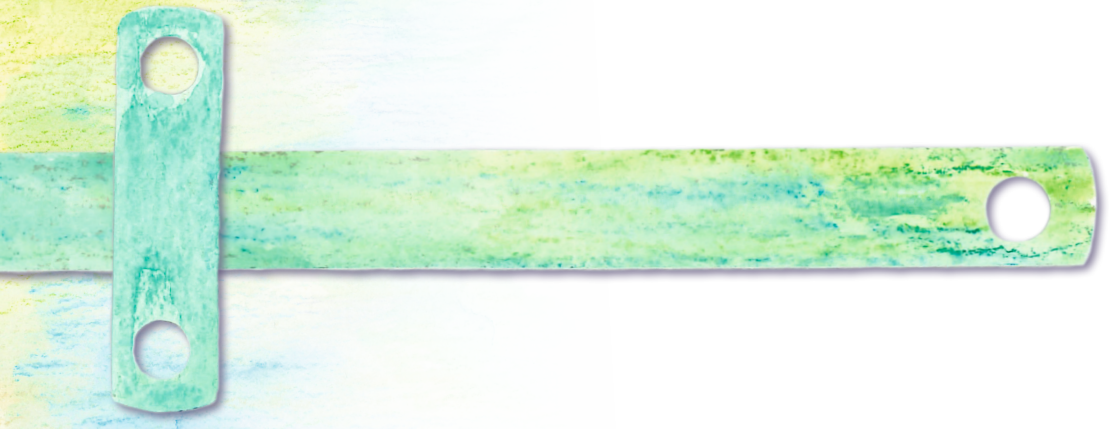
19. Emil S, Sévigny M, Montpetit K, et al. Success and duration of dynamic bracing for pectus carinatum: A four-year prospective study. J Pediatr Surg. 2017;52(1):124-9.

20. Port E, Hebal F, Hunter CJ, et al. Measuring the impact of brace intervention on pediatric pectus carinatum using white light scanning. J Pediatr Surg. 2018;53(12):2491-4.

21. Daemen JHT, Loonen TGJ, Lozekoot PWJ, et al. Optical imaging versus CT and plain radiography to quantify pectus severity: a systematic review and meta-analysis. J Thorac Dis. 2020;12(4):1475-87.
22. Brochhausen C, Turial S, Müller FK, et al. Pectus excavatum: history, hypotheses and treatment options. Interact Cardiovasc Thorac Surg. 2012;14(6):801-6.
23. Notrica DM, McMahon LE, Jaroszewski DE. Pectus Disorders: Excavatum, Carinatum and Arcuatum. Adv Pediatr. 2024;71(1):181-94.
24. Duncan Phillips J, Hoover JD. Chest Wall Deformities and Congenital Lung Lesions: What the General/Thoracic Surgeon Should Know. Surg Clin North Am. 2022;102(5):883-911.
25. Janssen N, Daemen JHT, van Polen EJ, et al. Pectus Excavatum: Consensus and Controversies in Clinical Practice. Ann Thorac Surg. 2023;116(1):191-9.
26. Scalise PN, Demehri FR. The management of pectus excavatum in pediatric patients: a narrative review. Transl Pediatr. 2023;12(2):208-20.
27. Pilegaard HK. Single centre experience on short bar technique for pectus excavatum. Ann Cardiothorac Surg. 2016;5(5):450-5.
28. Haecker FM, Sesia S. Non-surgical treatment of pectus excavatum. J Vis Surg. 2016;2:63.
29. Daemen JHT, de Loos ER, Vissers YLJ, et al. Intercostal nerve cryoablation versus thoracic epidural for postoperative analgesia following pectus excavatum repair: a systematic review and meta-analysis. Interact Cardiovasc Thorac Surg. 2020;31(4):486-98.
30. Cheng YL, Lin CT, Wang HB, Chang H. Pleural effusion complicating after Nuss procedure for pectus excavatum. Ann Thorac Cardiovasc Surg. 2014;20(1):6-11.
31. Sayan B, Bekiroglu N, Yuksel M. Pectus cross bars increase hospital readmission rates due to serous pleural effusion. Gen Thorac Cardiovasc Surg. 2022;70(4):352-8.
32. Oka N, Masai K, Okubo Y, et al. Clinical impact of multiple pectus bars on surgical outcomes following pectus excavatum repair. Interdiscip Cardiovasc Thorac Surg. 2024;39(4).
33. Obermeyer RJ, Godbout E, Goretsky MJ, et al. Risk factors and management of Nuss bar infections in 1717 patients over 25years. J Pediatr Surg. 2016;51(1):154-8.
34. Obermeyer RJ, Cohen NS, Gaffar S, et al. Multivariate analysis of risk factors for Nuss bar infections: A single center study. J Pediatr Surg. 2018;53(6):1226-9.
35. Ortega G, Rhee DS, Papandria DJ, et al. An evaluation of surgical site infections by wound classification system using the ACS-NSQIP. J Surg Res. 2012;174(1):33-8.
36. Bouchard ME, Petrosyan M, Kane TD. Case series of metal allergy following Nuss procedure: Not only for stainless steel bars. J Pediatr Surg. 2021;56(11):1976-81.
37. Abdellaoui S, Scalabre A, Piolat C, et al. Pectus Arcuatum: A Pectus Unlike Any Other. J Pediatr Surg. 2023;58(9):1679-85.

Part I

Pectus carinatum



Chapter 2

Ravitch surgery or dynamic compression bracing for pectus carinatum: a retrospective cohort study

Hendrik van Braak, Sjoerd A. de Beer, Sander Zwaveling, Matthijs W.N. Oomen, Justin R. de Jong

Annals of Thoracic Surgery. 2024 Jan 117:144-150.

ABSTRACT

Background

Pectus carinatum is a pediatric condition which can be treated by dynamic compression system (DCS) bracing or surgery. Several publications concerning DCS-bracing or surgery are published, however without comparing both treatments.

Methods

Over a 10 year period 738 patients with pectus carinatum were treated. We describe our 10-years' experience and results of both treatments.

Results

Of the 631 patients who received DCS-bracing treatment, 553 finished treatment and 78 patients are still under treatment. Seventy-three point eight percent (408/553) of patients finished treatment successfully, 13.6% (75/553) failed treatment and 12.7% (70/553) were lost to follow-up. Success rate decreased with an increasing pressure of initial compression (84.2%-67.3%). Marfan and Poland syndrome were related to bad results.

Ravitch surgery was performed in 105 patients with a success rate of 92.4%. Complications occurred in 32.4% of patients and 6.7% of patients had complications for which surgery was needed. No relationship was found between osteotomy or sternal fixation and outcomes or complications. Abramson procedure was performed in two patients.

Conclusions

DCS-bracing should be the treatment of choice in patients with pectus carinatum because its non-invasiveness, good results and a lower complication rate compared to surgery. Besides pressure of initial correction, motivation is an important factor influencing outcomes and compliance remains a major challenge in treating pectus carinatum using DCS-bracing. Bracing patients before their growth spurt should be discouraged. Patients with a higher pressure of initial compression (>8.0-8.5) and Marfan and Poland syndrome have poorer outcomes. In those cases surgery may be considered.

INTRODUCTION

Pectus carinatum (PC) is a pediatric condition characterized by overgrowth of costal cartilage which causes protrusion of the sternum and adjacent cartilage. PC has an incidence of 0.3-0.7% and affects males four times more frequently than females.[1,2] PC is highly associated with a disturbed body image, embarrassment and a reduced quality of life.[3] Patients are mostly asymptomatic and treatment is started mainly for aesthetic reasons. Patients that are symptomatic complain of pain, dyspnea, tachypnea with exertion and reduced endurance.[4]

Traditionally PC patients were treated surgically using the Ravitch procedure which was first introduced in 1960 and modified during later years.[5, 6] Another surgical technique is the Abramson procedure, which is a 'reverse' Nuss procedure.[7] From the seventies onwards orthotic treatment was introduced as a non-operative treatment with a success rate ranging from 40-90%. Orthotic treatment was complicated by long treatment time (up to 30 months) and complications such as discomfort, pain, skin rash or discoloration and skin lesions.[8-12] In 2008 Martinez-Ferro et al. introduced the dynamic compression system (DCS) for treating pectus carinatum and claimed a success rate up to 90%.[13, 14] This system was adopted in the Amsterdam Pectus Center in 2013. Several publications concerning the outcomes, complications and improvement of quality of life after DCS-bracing or Ravitch surgery were published, however most studies focused on either Ravitch [6, 15-18] or DCS treatment[1, 2, 19-23] without comparing both treatments.

Recently the first comparison of both treatments in one medical center was published.[24] In this study we describe the results of both the surgical procedures and DCS-bracing for patients with PC in our center.

PATIENTS AND METHODS

Study design

We conducted a retrospective cohort study including all patients with PC, aged 0-18 years (y), that were treated from 2009 to 2019 with either surgery (Ravitch or Abramson) or DCS-bracing. A Medical Research Ethics Committee (METC) official waiver of ethical approval was granted from the METC of the Amsterdam Medical Center. Informed consent for this specific study was prospectively obtained from all individual patients (or their parents) included in this study. Patients who were retrospectively included were contacted by their treating clinician to receive informed consent. This was recorded in their electronical medical record system. No patient opted out of providing consent.

Data collection

Patients were prospectively included, data were retrospectively complemented and reviewed using the patients electronical medical record system. Sixteen patients, who underwent Ravitch surgery before we started including patients prospectively for this study, were retrospectively included. Baseline characteristics were age, gender, breast growth, underlying diseases, symmetry and degree of the deformity, flaring, pressure of initial compression (PIC), symptoms, follow-up (months (m)), hospitalization (days (d)), details about the surgical procedure (osteotomy, fixation methods), length of treatment and prior surgeries. DCS-bracing patients were distributed into two age groups (<10y & 10-18y) and three groups based on PIC (<5.0 PSI (pounds per square inch), 5.0-7.5 PSI & >7.5 PSI). Patients were compared on different variables (PIC, age) to identify possible factors influencing the outcome of treatment.

Indications

After anamnesis and physical examination the type of treatment was determined using shared decision making. DCS-bracing was advised for all patients except for patients with pectus arcuatum (PA), an asymmetrical deformity, a high PIC (>10 PSI) or a history of earlier DCS-bracing. They were advised to choose Abramson or Ravitch surgery, but they could still choose DCS-bracing. Patients who were not eligible for DCS-bracing usually had a rigid, stiff thorax with a high PIC, a asymmetric deformity or PA. In these cases Abramson surgery is unfit because of more complications and the risk of plates breaking out.[25] Another reason for choosing Ravitch over Abramson was the fact that Ravitch procedure required only one surgery.

Techniques

For DCS-bracing the FMF Dynamic Compression System (FMF-DCS) was used (Figure 1). First we measure the PIC using a Pressure Measuring Device which is pressed on the chest and measures the average applied pressure until the deformity is corrected. Thereafter the thorax is measured, the brace is ordered, fitted and the patient receives training on how to use it. The brace is typically worn daily for 12-24 months, 12 hours a day, until the deformity is corrected. More about the procedures and technique can be found in our earlier manuscript[17] and a step-by-step plan on how to fit the brace is described by Martinez-Ferro et al.[14] Ravitch surgery was performed using the modified Ravitch technique as described by Welch et al.[6] Abramson surgery was performed using the technique as described by Abramson et al.[7]

Statistical analysis

Due to the nature of this study only descriptive measurements were used. Probability values of less than 0.05 were considered statistically significant. All data were analyzed using IBM SPSS Statistics 24.

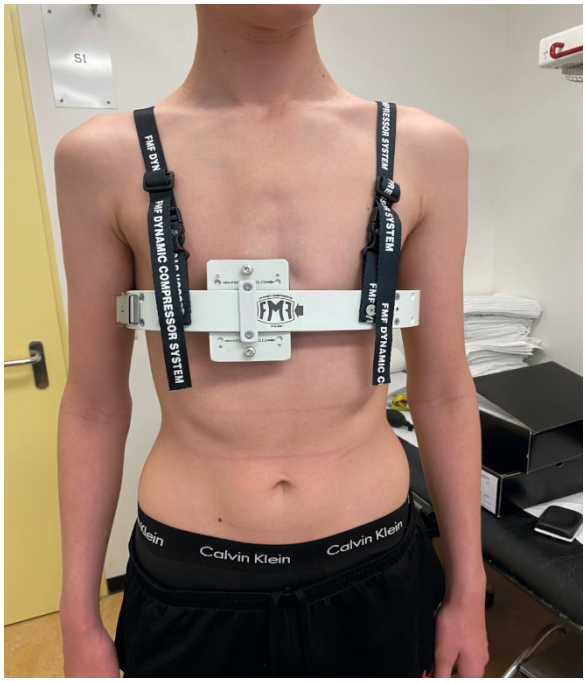


Figure 1 Patient wearing a Dynamic Compression System Brace

Outcomes

Primary outcomes were success or failure and complications (number of patients with complications and severity of complications). Active treatment as outcome was only possible for patients who received DCS-bracing. Treatment was regarded successful if patients achieved a satisfactory result, in consultation with the clinician. Pictures of the thorax were taken before and after treatment by a medical photographer to give patients and clinicians a reference. Failure for DCS-bracing was defined as: patients who started brace therapy but did not accomplish a sufficient result or could not endure bracing therapy. Patients were regarded lost to follow-up if they were not seen for two years in the outpatient clinic before ending treatment. Relapse (or failure) in patients who underwent surgery was defined as a disturbing comeback of PC after initial correction. Complications (wound infection, wound dehiscence, bleeding, pneumonia, pneumothorax, atelectasis, pseudoarthrosis, malunion, pectus excavatum (PE), painful/

numb/hypertrophic scar, pressure ulcers and dermatosis) were ranked on severity and impact using the Clavien-Dindo Classification.[26]

RESULTS

Baseline characteristics of all patients

Baseline characteristics can be found in Table 1. Between April 2009 and November 2019 738 patients were treated at the Amsterdam Pectus Center. Two patients deliberately opted for the Abramson procedure. Six hundred thirty-one patients started with DCS-bracing. One hundred and five patients applied for Ravitch surgery. Seven patients had a pectus deformity of the chondromanubrial (PA) type. Asymmetrical deformities, scoliosis, PA and deformities accompanied by costal flaring were found relatively more often in patients receiving Ravitch surgery. A little less than 50% of the patients was bullied or felt embarrassed about their chest (361/738).

Table 1 Patient characteristics and treatment groups

	n	%	Age [a] (y)	Follow-up [a] (m)
Total treated patients	738	100.0	14.0 (13.0-16.0)	53.0 (35.0-74.0)
Male	683	92.5		
Female	55	7.5		
Treatment type				
DCS	631	85.5	14.0 (13.0-15.0)	53.0 (35.0-71.0)
Ravitch	105	14.2	15.0 (14.0-16.0)	54.0 (34.0-79.0)
Abramson	2	0.3	15.5 (15.0-16.0)	85.0 (85.0-85.0)
Underlying conditions				
Marfan syndrome	10	1.4		
Poland syndrome	2	0.3		
Osteogenesis imperfecta type 3	1	0.1		
Scheuermann's disease	1	0.1		

Characteristics of deformities			**%Ravitch**	**%DCS**	**%Abramson**
Asymmetric	277	37.5	*65.7*	*32.6*	*100.0*
Symptomatic	276	37.4	*33.0*	*38.0*	*50.0*
Flaring	134	18.2	*21.7*	*17.4*	*50.0*
Scoliosis	92	12.5	*17.9*	*11.5*	*0.0*
Chondromanubrial (PA)	7	0.9	*6.6*	*0.0*	*0.0*

DCS = dynamic compression bracing, IQR = interquartile range, m = months, n = number of patients, PA = pectus arcuatum, PC = pectus carinatum, y = years
[a] Continuous variables expressed as median (IQR)

Outcomes of DCS-bracing

Figure 2 summarizes the DCS-bracing treatment group. Five hundred fifty three (87.6%) of patients had completed treatment. From those patients 73.8% (408/553) finished treatment successfully, 13.6% (75/553) failed and 12.7% (70/553) were lost to follow-up. The combination of not wearing the brace enough and a lack of motivation was (nearly) always the reason for failing treatment. Female patients (44/553) had a success percentage of 81.8% compared to 73.1% in male patients (509/553). No significant relationship was found between breast growth and outcome of DCS-bracing (P=0.64).

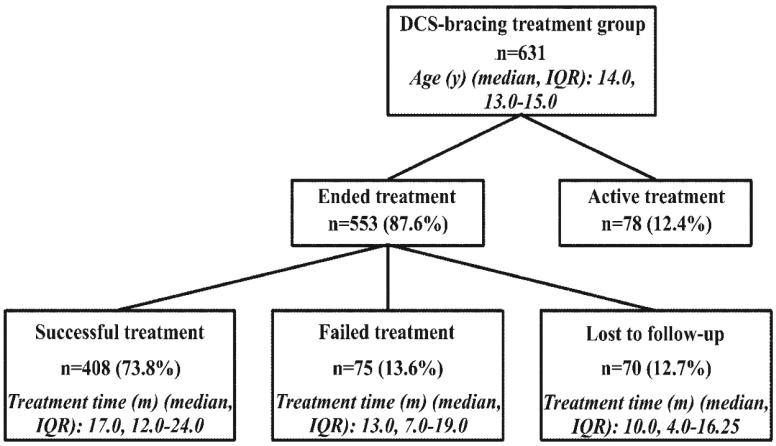


Figure 2 DCS-bracing group details (DCS = dynamic compression system, IQR = interquartile range, m = months, n = number of patients, y = years)

As earlier research suggested that in patients with a higher PIC treatment is less successful,[13] patients were divided into three PIC-groups. Results are shown in Table 2. The PIC varied from 1.9 to 13.3 PSI. Patients with a PIC less than 5.0 PSI had the best results with 84.2% finishing treatment successfully. Higher PIC decreased the rate of success, with success percentages decreasing to 60.0% when the PIC exceeded 10.0 PSI (Figure 3). A statistically significant (P=0.01) inversely proportional relation was found between success of DCS-bracing and PIC. A higher PIC was also related with longer treatment duration and higher age (respectively P<.001 & P<.001). No relevant complications of DCS-bracing were noted. Younger patients (<10y) had initially better results (83.9% treated successfully) compared to other age groups (10-18y: 73.2% treated successfully), but no statistical relation was found between age and outcomes (P=0.59), recurrence (P=0.74) or length of retainer mode trajectory (P=0.48). Four percent of the patients suffered from minor skin lesions during the treatment. One patient (0.2%) had overcorrection and developed PE for which the patient was unsuccessfully treated with vacuum bell therapy. The patient subsequently received a

Nuss-bar. Twenty-three patients (3.6%) decided to quit bracing therapy and received a Ravitch procedure.

Table 2 Pressure of initial correction (PIC) & results

	n	%	Treatment time [a] (m)
Total treated	631	100.0	
PIC < 5.0	116	18.4	
PIC 5.0-7.5	275	43.6	
PIC > 7.5	240	38.0	
PIC < 5.0 (active patients left out)			
Successful	80	84.2	15.0 (11.0-19.0)
Failed	7	7.4	
Lost to follow-up	8	8.4	
PIC 5.0-7.5 (active patients left out)			
Successful	184	75.4	16.0 (12.0-23.0)
Failed	29	11.9	
Lost to follow-up	31	12.7	
PIC > 7.5 (active patients left out)			
Successful	144	67.3	19.5 (14.5-25.0)
Failed	39	18.2	
Lost to follow-up	31	14.5	

IQR = interquartile range, m = months, n = number of patients, PIC = pressure of initial correction
[a] Continuous variables expressed as median (IQR)

Table 3 Ravitch treatment group details

	n	%
Ravitch patients never braced	94	89.5
Ravitch after DCS failure	9	8.6
Ravitch after Nuss	2	1.9
Sternal fixation after osteotomy		
None	47	61.0
Soluble stitches	15	19.5
Non-soluble stitches	4	5.2
LCP-plate	11	14.3
Relapse PC	8	7.6

DCS = dynamic compression bracing, n = number of patients, PC = pectus carinatum

Outcomes of Ravitch/Abramson surgery

Table 3 summarizes the Ravitch treatment bracing group. Mean hospitalization time was 6.4 days (SD=1.3). Surgery was performed on patients who did not receive prior brace therapy (94/105, 89.5%), failed DCS-bracing (9/105, 8.6%), or were treated with a Nuss-bar before and needed treatment for overcorrection (2/105, 1.9%). The PIC of patients who failed DCS-bracing was high with an median of 9.0 PSI (IQR 7.8-9.2), compared to 7.0 PSI (IQR 5.6-8.0) in the total DCS-bracing population. Ninety-seven (92.4%) of the patients finished treatment with satisfying results. Relapse occurred in eight patients (7.6%). Sixteen patients received Ravitch surgery before the introduction of DCS-bracing. The success rate of surgery was lower before (77.5%) than after the introduction (93.3%) with a comparable percentage of complications (respectively 31.3% and 32.6%).

Complications of Ravitch surgery are summarized in Table 4. Thirty-four patients (32.4%) had complications. There were seventeen patients with one complication, thirteen patients with two complications and four patients with three complications or more. Patients with complications spent on average 1.0 days longer in the hospital compared to patients without complications (P=0.01). Grade IIIb complications, for which an intervention under general anesthesia is needed, were pseudoarthrosis (5), wound dehiscence (1) and hypertrophic scars (1). Eight patients developed PE, for which three patients were successfully treated with a vacuum bell. No statistically relevant relations were found between osteotomy and the amount of complications (P= 0.92) nor the severity of the complications (P=0.67). No statistical relevant relation was found between PE and sternal fixation method (P=0.11). Relapse was not related to osteotomy nor complications (respectively P=0.91 & P=0.38). Two patients successfully underwent Abramson surgery, one patient needed an additional procedure for resection of flaring. No complications were noted.

DISCUSSION

In general, both DCS-bracing and surgery obtained good results. In the DCS-bracing group the success rate decreased with an increasing PIC, with a steep decrease in success percentage when the PIC exceeds 8.0-8.5. This, in combination with the high median PIC (9.0 PSI) in patients who underwent Ravitch surgery after failing DCS-bracing, makes DCS-bracing in patients with a high PIC debatable. Figure 3 however shows that the success rate increases slightly after reaching its ultimate low at a PIC of 9.0-9.5. A reason for this could be that the patients with a high PIC deliberately made a highly motivated choice for DCS-bracing, despite being well informed about the difficulty of DCS-bracing in their situation by their attending surgeon. This, combined

with the fact that overall a lack of motivation is the most important reason for DCS-bracing failure, might prove that not primarily the PIC, but also motivation is one of the most important factors determining the success of DCS-bracing, which might prove to be clinical useful.

Table 4 Complications of Ravitch repair

	n	**%**
Complications	56	100.0
Hypertrophic scar	14	25.0
Pectus excavatum	8	14.3
Painful scar	7	12.5
Pseudoarthrosis	5	8.9
Wound infection	5	8.9
Numb scar	3	5.4
Pneumothorax	3	5.4
Wound dehiscence	3	5.4
Malunion	2	3.6
Bleeding	2	3.6
Atelectasis	1	1.8
Dermatosis	1	1.8
Pneumothorax	1	1.8
Pressure ulcers	1	1.8
Clavien-Dindo Classification		
I	37	66.1
II	12	21.4
IIIb	7	12.5

n = number of patients

Bracing in the young patients

It was expected that young patients (36) would struggle with a prolonged time of wearing the brace and relapse. Yet within the study period no recurrence occurred and no differences in retainer mode time were found between age groups. However secondary evaluation showed that, several years after the study period, eight (22.2%) patients contacted the clinic again because of relapse. Those patients restarted treatment. More than half of the other patients wear their brace occasionally at night to maintain the obtained results. Relapse occurred nearly always in their growth spurt. DCS-bracing should probably be delayed for several years until children have had their growth spurt.

DCS-bracing in the literature

When DCS-bracing was first adopted great results were expected. In the original article of Martinez-Ferro et al. a success rate of 88.4% was mentioned[13] while others noted a success rate of 100%.[19, 23] Those studies were, however, a lot smaller and Martinez-Ferro et al. excluded patients with a PIC above 10.0 PSI. Our study included 38 patients with a PIC of 10.0 PSI or higher. Their results were also influenced by the exclusion of 24 (10.3% of the total population) patients with Marfan or Poland syndrome. Both syndromes seem to be related to bad outcomes, which could have raised the success rate in their study. This is confirmed by our results. In our study two (33.3%) patients with either Marfan or Poland syndrome finished DCS-bracing successfully, three were lost to follow-up and one failed treatment. In patients with Marfan or Poland syndrome surgery could be considered the treatment of first choice.

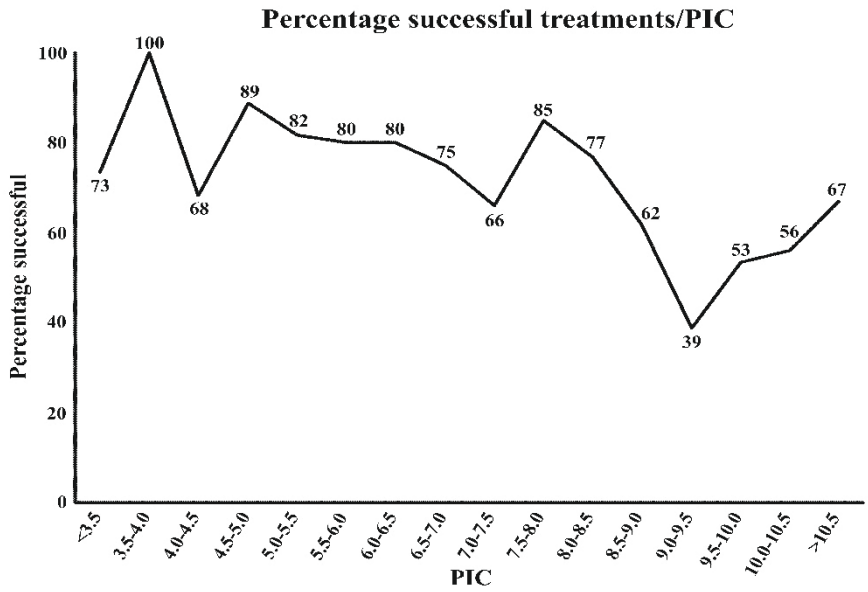


Figure 3 Percentage of successful DCS-treatment per PIC (DCS = dynamic compression system, PIC = pressure of initial correction)

In recent literature Kelly et al. et al. also evaluated the outcomes of DCS-bracing of the last ten years. They showed a much lower success percentage of 50.2% with a loss to follow-up of 41.1%.[24] In our study 12.7% of the patients were lost to follow-up, which is in accordance with the 13.4% noted by Martinez-Ferro et al. Despite this, compliance remains a major problem of the DCS-bracing. There is room for improvement if patients could stay motivated. Earlier studies stated social discomfort and lack of motivation as major contributors to decreased compliance and loss of follow-up.[2, 13, 21] Monitoring (non-)compliance, for example with a real-time monitoring system such as MyPectus[27], could be useful for early detection of non-compliance and could give more insight in the reasons behind it. Kelly et al. experimented with an optical scanning device to monitor the progress of the chest deformity. This could be shown to patients to prove the efficacy of the therapy and encourage them to continue wearing it.[28] Another study did the same with a white light scanner.[29] The use of the Pectus Carinatum Body Image Quality of Life questionnaire (PeCBI-QOL) could also help improve and monitor the body image of patients.[30]

Ravitch and Abramson surgery

Ravitch surgery received excellent outcomes with 92.4% of the patients successfully treated compared to 73.8% in the DCS-bracing group. Despite this, surgery leads to more complications. While no major complications were observed in the bracing group, one in three surgical patients had postoperative complications. Most complications were minor and only 6.7% of the patients with complications needed surgery under general anesthesia. This corresponds with other studies on Ravitch surgery.[15, 18] The results are also similar to recent figures on the Abramson procedure: satisfactory results in 91.0% of patients and complications in 26.5% of patients (with 6.6% of patients needing surgery because of complications)[25]. Despite the Abramson procedure being a minimal invasive technique with only little scars, patients in our study who were suitable for this procedure all received a DCS-brace, except for one patient who preferred the Abramson procedure over DCS-bracing. Patients who were not eligible for bracing were also unfit for Abramson (asymmetric, flaring, high PIC) and therefore underwent Ravitch surgery except for one patient. This patient, despite having a asymmetrical thorax with costal flaring, deliberately opted for Abramson surgery. This patient needed additional surgery to remove the costal flaring which could not be corrected using the Abramson procedure.

Conclusion

In our opinion, DCS-bracing should be the treatment of first choice in patients with PC. With a success rate varying from 67.3-84.2% results are not as high as the percentages achieved by surgery, but the risks are much lower and if conservative therapy fails there is still the possibility to opt for surgery. Bracing patients before their growth spurt should be discouraged, unless they are willing and motivated to wear the brace in retainer mode for many years. Besides PIC, motivation seems to be an important factor influencing the outcomes. If compliance is suspected to be a problem or patients have complex deformities (PA, underlying syndromes like Marfan or Poland, asymmetry, and flaring) primary surgery should be considered.

Limitations

This study might seem a two-era study, before and after the introduction of DCS-bracing in 2013. Before 2013 only patients with major deformities applied for treatment. While the indication for surgery remained the same after 2013, some patients with less severe deformities, despite being advised to choose DCS-bracing, wanted surgery too. Perhaps this explains the differences in success rate of Ravitch surgery before and after 2013. Although only 16 patients were treated before 2013, this could bias the results.

It is difficult to quantify both degree of deformity before treatment and outcomes after treatment in Ravitch and DCS-bracing patients since PC is mainly a cosmetic problem. Although some clinics use 3D imaging to map the deformities this might not be the ultimate solution, since the appearance of the deformity also depends on muscle mass and BMI. The quantification of initial deformities and outcomes remains a hurdle which is yet to be taken. Further investigation into the reasons for discontinuing DCS-bracing and loss of follow-up is needed to optimize DCS-bracing. No evaluation of resolution of symptoms in symptomatic patients was performed in this study because we lack data about the resolution of symptoms after treatment.

REFERENCES

1 De Beer SA, Blom YE, Lopez M, De Jong JR. Measured dynamic compression for pectus carinatum: a systematic review. Semin Pediatr Surg. 2018;27(3):175-182.

2 De Beer SA, Gritter M, De Jong JR, Van Heurn ELW. The dynamic compression brace for pectus carinatum: intermediate results in 286 patients. Ann Thorac Surg. 2017;103(6):1742-1749.

3 Desmarais TJ, Keller MS. Pectus carinatum. Curr Opin Pediatr. 2013;25(3):375-381.

4 McHam B, Winkler L. Pectus carinatum. StatPearls. StatPearls Publishing Copyright © 2020, StatPearls Publishing LLC.; 2020.

5 Ravitch MM. Operative correction of pectus carinatum (pigeon breast). Ann Surg. 1960;151(5):705-714.

6 Welch KJ, Vos A. Surgical correction of pectus carinatum (pigeon breast). J Pediatr Surg. 1973;8(5):659-667.

7 Abramson H, Aragone X, Blanco JB, Ciano A, Abramson L. Minimally invasive repair of pectus carinatum and how to deal with complications. J Vis Surg. 2016;2:64.

8 Wong KE, Gorton GE 3rd, Tashjian DB, Tirabassi MV, Moriarty KP. Evaluation of the treatment of pectus carinatum with compressive orthotic bracing using three dimensional body scans. J Pediatr Surg. 2014;49(6):924-927.

9 Loff S, Sauter H, Wirth T, Otte R. Highly efficient conservative treatment of pectus carinatum in compliant patients. Eur J Pediatr Surg. 2015;25(5):421-424.

10 Lee RT, Moorman S, Schneider M, Sigalet DL. Bracing is an effective therapy for pectus carinatum: interim results. J Pediatr Surg. 2013;48(1):184-190.

11 Colozza S, Bütter A. Bracing in pediatric patients with pectus carinatum is effective and improves quality of life. J Pediatr Surg. 2013;48(5):1055-1059.

12 Frey AS, Garcia VF, Brown RL, et al. Nonoperative management of pectus carinatum. J Pediatr Surg. 2006;41(1):40-45.

13 Martinez-Ferro M, Fraire C, Bernard S. Dynamic compression system for the correction of pectus carinatum. Semin Pediatr Surg. 2008;17(3):194-200.

14 Martinez-Ferro M, Bellia-Munzon G, Fraire C, et al. Non-surgical treatment of pectus carinatum with the FMF(®) Dynamic Compressor System. J Vis Surg. 2016;2:57.

15 Davis JT, Weinstein S. Repair of the pectus deformity: results of the Ravitch approach in the current era. Ann Thorac Surg. 2004;78(2):421-426.

16 Pilegaard H, Licht PB. Minimal invasive repair of pectus excavatum and carinatum. Thorac Surg Clin. 2017;27(2):123-131.

17 Emil S. Current options for the treatment of pectus carinatum: when to brace and when to operate? Eur J Pediatr Surg. 2018;28(4):347-354.

18 Del Frari B, Sigl S, Schwabegger AH. Complications related to pectus carinatum correction: lessons learned from 15 years' experience. Management and literature review. Plast Reconstr Surg. 2016;138(2):317-329.

19 Lopez M, Patoir A, Varlet F, et al. Preliminary study of efficacy of dynamic compression system in the correction of typical pectus carinatum. Eur J Cardiothorac Surg. 2013;44(5):316-319.

20 Cohee AS, Lin JR, Frantz FW, Kelly Jr. RE. Staged management of pectus carinatum. J Pediatr Surg. 2013;48(2):315-320.

21 Dekonenko C, Dorman RM, Pierce A, et al. Outcomes following dynamic compression bracing for pectus carinatum. J Laparoendosc Adv Surg Tech A. 2019;29(10):1223-1227.I

22 Poola AS, Pierce AL, Orrick BA, et al. A single-center experience with dynamic compression bracing for children with pectus carinatum. Eur J Pediatr Surg. 2018;28(1):12-17.

23 Sesia SB, Holland-Cunz S, Häcker FM. Dynamic compression system: an effective nonoperative treatment for pectus carinatum: a single center experience in Basel, Switserland. Eur J Pediatr Surg. 2016;26(6):481-486.

24 Kelly Jr. RE, Obermeyer RJ, Goretsky MJ, et al. Ten-year experience with staged management of pectus carinatum: results and lessons learned. J Pediatr Surg. 2021;56(10):1835-1840.

25 Geraedts TCM, Daemen JHT, Vissers YLJ, et al. Minimally invasive repair of pectus carinatum by the Abramson method: A systematic review. J Pediatr Surg. 2021;S0022-3468(21)00829-0.

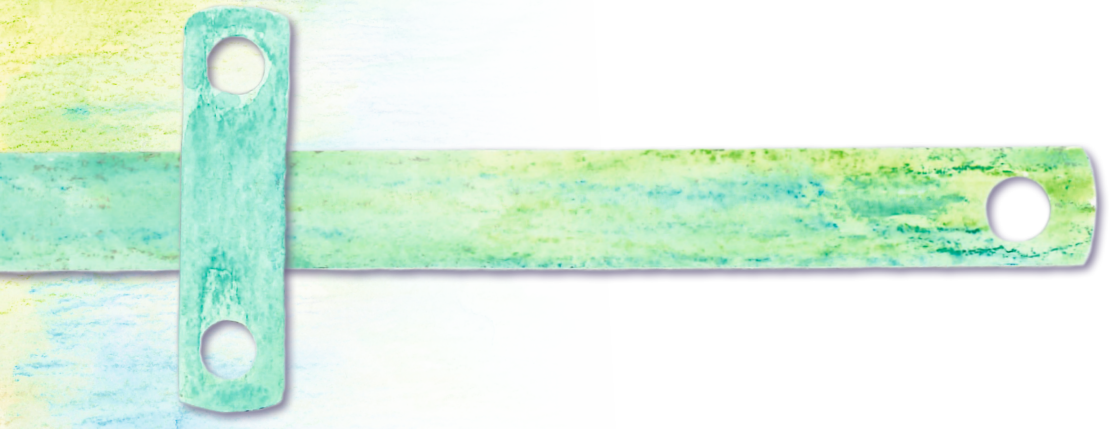
26 Clavien PA, Barkun J, De Oliveira ML, et al. The Clavien-Dindo classification of surgical complications: five-year experience. Ann Surg. 2009;250(2):187-196.

27 Harrison B, Stern L, Chung P, et al. First-in-human pilot study of remote compliance monitoring of teens using dynamic compression bracing to correct pectus carinatum. J Pediatr Surg. 2016;51(4):608-611.

28 Kelly Jr. RE, Obermeyer RJ, Kuhn MA, et al. Use of an optical scanning device to monitor the progress of noninvasive treatments for chest wall deformity: a pilot study. Korean J Thorac Cardiovasc Surg. 2018;51(6):390-394.

29 Port E, Hebal F, Hunter CJ, Malas B, Reynolds M. Measuring the impact of brace intervention on pediatric pectus carinatum using white light scanning. J Pediatr Surg. 2018;53(12):2491-2494.

30 Paulson JF, Ellis K, Obermeyer RJ, et al. Development and validation of the pectus carinatum body image quality of life (PeCBI-QCL) questionnaire. J Pediatr Surg. 2019;54(11):2257-2260.



Chapter 3

Improving quality of life with dynamic compression bracing in patients with pectus carinatum

Hendrik van Braak, Sjoerd A. de Beer, Jos W.R. Twisk, Sander Zwaveling, Matthijs W.N. Oomen, Justin R. de Jong, L.W. Ernest van Heurn

Journal of Pediatric Surgery. 2024 Jan 60:161975.

ABSTRACT

Background

Patients with pectus carinatum have lower quality of life and self-esteem than their peers. We assessed the impact of dynamic compression system bracing on quality of life in patients with pectus carinatum.

Methods

We conducted a prospective cohort study on patients aged 10-21 years. We assessed quality of life using the Child Health Questionnaire-87, the State-Trait Anxiety Inventory-6, the World Health Organization Quality of Life-BREF, the 36-Item Short Form Survey, and the Single-Step Questionnaire adapted for pectus carinatum.

Results

Between March 2013 and March 2016, 225 patients treated with dynamic compression system bracing were included. Patients showed improvements across the overall scores of the 36-Item Short Form Survey (Δ7.7 (2.9-12.4)), Single-Step Questionnaire (Δ4.1 (2.0-6.3)) and three out of four World Health Organization Quality of Life-BREF domains (physical health (Δ8.7 (3.7-13.7)), psychological health (Δ11.8 (6.1-17.5)), environment (Δ5.7 (0.2-11.3))). No changes across the Child Health Questionnaire-87 overall score were observed (Δ5.5 (-0.5-11.5)). Most improvement occurred within six to twelve months after treatment initiation, stabilizing thereafter. Anxiety scores on the State-Trait Anxiety Inventory-6 did not improve (Δ0.5 (-0.1-1.2)). Scores on physical complaints, pain, psychological health and self-esteem/self-image improved across all questionnaires. In contrast to the successfully treated group, the unsuccessfully treated group showed no improvement on any of the questionnaires. Most patients (87.2%) would choose bracing again, 94.9% of patients were satisfied with the treatment.

Conclusions

Dynamic compression system bracing improves quality of life, reduces physical complaints and pain and boosts psychological health and self-esteem in patients with pectus carinatum.

INTRODUCTION

Pectus carinatum (PC) is a pediatric condition marked by abnormal overgrowth of costal cartilage which results in protrusion of the sternum. Incidence varies from 0.04-0.7% in the current literature.[1,2] Almost 50% of the patients feel embarrassed about their chest and experience occasional bullying. This may lead them to avoid activities in which their chest is visible such as swimming. It may cause psychological distress, a disturbed body image and an overall reduced quality of life (QoL).[2-4] Additionally, patients often report various physical complaints, primarily respiratory and cardiovascular, but also pain.[5,6] Currently, dynamic compression system bracing (DCS-bracing) is the treatment option of first choice, with good results.[2] Despite these results, there is limited knowledge about the effect of DCS-bracing on improvements in QoL, self-esteem and psychological distress. Only three studies have addressed (facets of) QoL after treatment; however, they used different braces and bracing protocols.[7-9] Two studies report on improvement of QoL after surgical correction of PC.[10,11] The lack of studies on this topic is particularly surprising as a reduced quality of life is the primary reason for seeking treatment.

Therefore, we evaluate the changes in QoL in patients with PC treated using DCS-bracing, with emphasis on physical complaints, pain, psychological health and self-esteem/self-image.

PATIENTS AND METHODS

Patients and study design

We conducted a prospectively cohort study of patients who commenced DCS-bracing therapy between March 2013 and March 2016 at the Amsterdam Pectus Centre. Patients aged ten or older with sufficient knowledge of the Dutch language were included. A Medical Ethics Review Committee (METC) official waiver of ethical approval was granted by the METC of the Amsterdam Medical Center. Informed consent was obtained from all patients (or their parents) included in this study.

Treatment protocol

At the Amsterdam Pectus Centre, patients with pectus carinatum (PC) are treated using the FMF Dynamic Compression System brace (Pampamed). Pediatric surgeons handle both intake and follow-up appointments, following the treatment protocol outlined in Figure 1. Patients are advised to delay treatment until after the onset of their growth spurt. The pressure of initial correction is assessed using a pressure device that measures the average force required to correct the chest deformity. Patients are advised to wear the brace as much as possible, and we encourage them to wear the

brace at night to extend their treatment hours more comfortably. Treatment is deemed successful when a satisfactory outcome is achieved, as determined in consultation with the clinician.

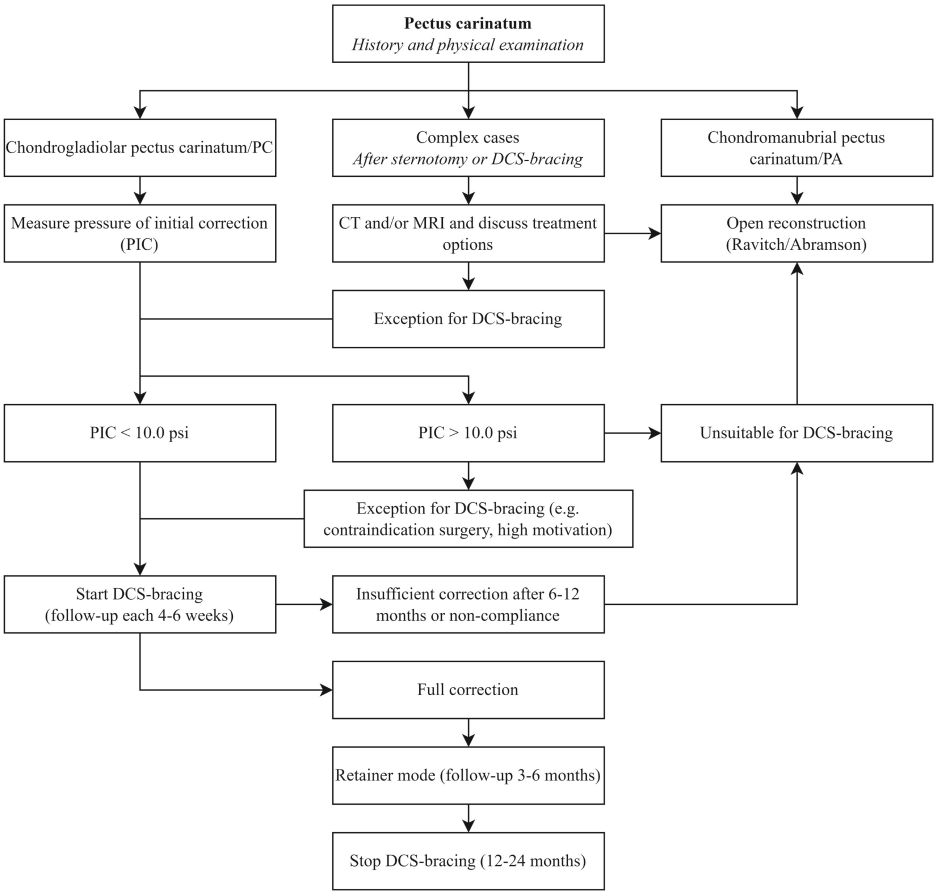


Figure 1 Treatment protocol DCS-bracing (CT = Computer Tomography, DCS = Dynamic Compression System, MRI = Magnetic Resonance Imaging, PA = Pectus Arcuatum, PC Pectus = Carinatum, PIC = Pressure of Initial Correction)

Questionnaires

Patients were divided into three age groups (10-16y, 16-18y and >18y), to meet the validation criteria for the various questionnaires, with distinct questionnaires administered to each age group. Patients younger than ten years were excluded. Questionnaires were administered at the start of treatment and at one, six, twelve, eighteen and 24 months thereafter.

The Single Step Questionnaire (SSQ) (Appendix A), originally developed to assess QoL in patients with PE after the Nuss procedure, was used to assess change in QoL and satisfaction during treatment.[12] The questionnaire was adapted in our center to fit PC patients and was used with all subjects. All items, except for three, were scored on a 5-point Likert scale.

Anxiety was assessed using the Six-item State-Trait Anxiety Inventory (STAI-6) (Appendix B).[13] Items were scored on a 4-point Likert scale. The total score was displayed on a 0-24 scale.

The evaluation of QoL in the younger patients, aged ten to eighteen years, was done with the Child Health Questionnaire-87 (CHQ-87) (Appendix C). The CHQ-87 encompasses different domains, covering physical, emotional, and social well-being domains, including specific inquiries regarding self-esteem and self-image perception.[14] Items were scored using a 4- or 5-point Likert scale. Domain scores were converted to a standardized 0-100 scale.

The World Health Organization Quality of Life Brief Version (WHOQOL-BREF) (Appendix D), a condensed version of the World Health Organization Quality of Life-100 (WHOQOL-100), was used to assess QoL in patients aged sixteen years and older across four key domains, being physical health, psychological health, social relationships and environment. Additionally, it assesses two overarching aspects (QoL and health satisfaction in general). To enhance our study's scope, we added the facet body image and facet pain of the WHOQOL-100. Items were scored on a 5-point Likert scale. Domain scores were converted to a standardized 0-100 scale.[15]

Besides the WHOQOL-BREF the Short Form Survey 36 (SF-36) (Appendix E), a 36-item questionnaire covering eight domains of health, was used for patients aged sixteen years and older. This questionnaire also focusses on the limitations in daily life due to physical health or emotional problems.[16] We calculated the physical and mental component scores (PCS and MCS) besides the total score of the SF-36 to measure

QoL, as previously advised.[17,18] Items were scored on a combination of Likert scales and binary scales. Domain scores were converted to a standardized 0-100 scale.

Scores of all items in the questionnaires were converted to positive scores to standardize the scoring. Higher scores indicated a better QoL.

Statistical analysis

Data were analyzed using IBM SPSS Statistics 28.0. Descriptive measurements were utilized to characterize the study population. Changes in QoL primary and secondary outcomes were analyzed using linear mixed models, given that this method is ideal for analyzing repeated measures with missing data.[19] The linear mixed models only included time, which was treated as a categorical outcome variable represented by five dummy variables.

We used 95% Confidence Intervals (95% CI) to determine statistical significance. A 95% Confidence Interval that does not include the null value indicates that the result is statistically significant at the P<.05 level. The change in questionnaire scores is reported in the text as the absolute difference with 95% Confidence Intervals, formatted as (absolute difference (95% CI)), e.g., (Δ5.5 (-0.5-11.5)).

Outcomes

Primary outcomes were changes in QoL due to DCS-bracing. Secondary outcomes were changes in anxiety scores, changes in subdomains of the questionnaires, especially physical complains, pain, psychological health and self-esteem/self-image, and differences in outcomes between successfully and unsuccessfully treated patients.

RESULTS

Of all patients (N=260) treated in the study period 225 patients were included. Thirty-five patients were excluded either because of insufficient knowledge of the Dutch language, or they did not return any of the questionnaires. Baseline characteristics are listed in Table 1. Of all patients, 92.0% were male (207/225), median age was 15.0 (interquartile range (IQR) 13.0-16.0) years and median treatment time was 16.0 (IQR 12.0-23.0) months. All patients were diagnosed with PC, and none with pectus arcuatum (also known as Type 2 PC or Currarino-Silverman syndrome). None of the patients were still actively treated at the time of evaluation, 78.2% (176/225) finished treatment successfully, 13.3% (30/225) finished treatment unsuccessfully and 8.4% (19/225) were lost to follow-up. The number of respondents for each questionnaire at each time

point is detailed in Table 2. Initial response rates ranged from 62.7% to 77.3% at the zero-month mark, with a gradual decline over time.

CHQ-87

Table 3 shows the absolute and relative changes in overall scores of all questionnaires, while Figure 2 visually represents these changes in a graph to provide a clear overview. Total scores on the CHQ-87 did not change over the two years follow-up (Δ5.5 (-0.5-11.5)), although it increased from 76.1 to 81.6 points. More than half of this improvement occurred within the first six months of treatment. There were changes in the following subdomains: physical functioning (Δ1.9 (0.3-3.6)), role/social limitations due to behavior (Δ2.2 (0.6-3.8)), pain (Δ6.3 (1.1-11.6)), mental health (Δ3.0 (0.1-5.9)), and self-esteem (Δ3.4 (0.2-6.4)). There were no changes in the subdomains general health (Δ3.9 (-1.0-8.8)), role/social limitations due to physical health (Δ1.9 (-0.0-3.8)), role/social limitations due to emotional health (Δ2.3 (-0.4-5.0)), behavior (Δ2.4 (-0.1-4.9)), family cohesion (Δ 16.9 (-1.5-35.2)), or family activities (Δ0.9 (-2.8-4.5)). The positive change in health increased over the two years from 3.2 to 7.1 points (Δ4.0 (2.3-5.6)).

Table 1 Baseline characteristics

	All patients (N=225)
Sex [b]	
Male	207 (92.0)
Female	18 (8.0)
Age (years) [a]	15.0 (13.0-16.0)
<16 years [b]	158 (70.2)
16-18 years [b]	60 (26.7)
>18 years [b]	7 (3.1)
Treatment time (months) [a]	16.0 (12.0-23.0)
Results [b]	
Treatment successful	176 (78.2)
Treatment not successful	30 (13.3)
Lost to follow-up	19 (8.4)

[a] Nonparametric variables; expressed as median (IQR)
[b] Data displayed as n (%)

Table 2 Responses per questionnaire

	0m [a]	1m [a]	6m [a]	12m [a]	18m [a]	24m [a]
CHQ-87 *(N=223 [b])*	161	123	73	69	30	4
SF-36 *(N=59 [b])*	38	26	20	6	8	12
WHOQOL-BREF *(N=59 [b])*	37	31	23	15	8	15
STAI-6 *(N=225 [b])*	174	127	85	76	43	52
SSQ *(N=225 [b])*		127	97	68	43	75

[a] n of responses
[b] Number of patients eligible for this questionnaire at 0 months
CHQ-87 = Child Health Questionnaire-87, m = months, N/n = number, SF-36 = Short Form Survey 36, SSQ = Single Step Questionnaire, STAI-6 = Six-item State-Trait Anxiety Inventory, WHOQOL-BREF = World Health Organization Quality of Life Brief Version

SF-36

Total scores on the SF-36 did change over two years (Δ7.7 (2.9-12.4)), increasing from 83.4 to 91.0 points. QoL improved until six months after treatment initiation and then remained stable without decline. The PCS (Δ0.5 (-4.4-3.4)) and the MCS (Δ1.3 (-5.6-3.0)) did not change over the treatment period. There were changes in the subdomains physical functioning (Δ4.4 (1.2-7.6)), role of limitations due to physical health (Δ11.2 (3.1-19.4)), energy/fatigue (Δ10.5 (4.2-16.8)), emotional well-being (Δ7.4 (2.2-12.7)), social functioning (Δ11.9 (5.9-17.9)), pain (Δ9.0 (3.5-14.4)), and general health (Δ10.5 (3.1-18.0)). There were no changes in the subdomain role of limitations due to emotional problems (Δ8.8 (-1.0-16.5)). The positive change in health was stable during the two years and did not change (Δ4.2 (-3.1-11.5)).

WHOQOL-BREF

On all domains most improvement was made in the first six months after treatment initiation, after that it remained stable. There were changes in the subdomains physical health (Δ8.7 (3.7-13.7)), psychological health (Δ11.8 (6.1-17.5)), environment (Δ5.7 (0.2-11.3)), pain (Δ13.1 (3.2-23.0)), body image (Δ37.7 (29.5-45.8)). General QoL (Δ10.9 (5.5-16.3)) and health satisfaction (Δ10.7 (4.3-17.1)) did also improve. There was no change in the subdomain social relationships (Δ1.2 (-4.5-6.9)).

STAI-6

Total scores on the STAI-06 did not change over two years (Δ0.5 (-0.1-1.2)), although it increased from 21.0 to 21.5 points over two years. Most improvement was made in the first six months, with a slight decrease in total scores after six months.

Table 3 Absolute and relative change in (overall) scores of questionnaires

	0m	1m	6m	12m	18m	24m
CHQ-87						
Mean [a]	76.1 (6.8)	76.6 (6.9)	79.3 (4.4)	79.6 (4.2)	79.1 (4.3)	81.6 (7.1)
Difference from baseline [b]		*0.5 (0.7)*	*3.2 (4.2)*	*3.5 (4.6)*	*3.0 (3.9)*	*5.5 (7.2)*
SF-36						
Mean [a]	83.4 (13.7)	85.6 (13.2)	91.3 (7.5)	92.7 (5.3)	89.8 (8.0)	91.0 (10.5)
Difference from baseline [b]		*2.2 (2.6)*	*7.9 (9.5)*	*9.3 (11.2)*	*6.4 (7.7)*	*7.7 (9.1)**
WHOQOL-BREF						
Physical health						
Mean [a]	78.4 (12.1)	79.4 (15.4)	87.1 (10.8)	87.4 (10.8)	86.8 (10.4)	87.1 (10.1)
Difference from baseline [b]		*1.0 (1.3)*	*8.7 (11.1)*	*9.0 (11.5)*	*8.4 (10.7)*	*8.7 (11.1)**
Psychological health						
Mean [a]	71.7 (13.5)	77.1 (14.8)	82.5 (11.7)	83.3 (9.5)	81.9 (9.2)	83.5 (9.3)
Difference from baseline [b]		*5.4 (7.5)*	*10.8 (15.1)*	*11.6 (16.2)*	*10.2 (14.2)*	*11.8 (16.5)**
Social relationships						
Mean [a]	79.8 (14.1)	78.2 (15.5)	82.2 (11.7)	81.4 (14.2)	83.9 (11.9)	81.0 (12.0)
Difference from baseline [b]		*-1.6 (-2.0)*	*2.4 (3.0)*	*1.6 (2.0)*	*4.1 (5.1)*	*1.2 (1.5)*
Environment						
Mean [a]	81.1 (10.3)	81.8 (13.0)	88.8 (10.6)	88.4 (8.9)	86.9 (11.6)	86.9 (9.3)
Difference from baseline [b]		*0.7 (0.9)*	*7.7 (9.5)*	*7.3 (9.0)*	*5.7 (7.1)*	*5.7 (7.1)**

Table 3 Absolute and relative change in (overall) scores of questionnaires *Continued.*

	0m	1m	6m	12m	18m	24m
STAI-6						
Mean [a]	21.0 (2.7)	21.3 (2.6)	21.9 (1.7)	21.8 (2.2)	21.2 (2.3)	21.5 (2.4)
Difference from baseline [b]		*0.3 (1.4)*	*1.0 (4.3)*	*0.8 (3.8)*	*0.3 (1.0)*	*0.5 (2.4)*
SSQ						
Mean [a]		68.6 (7.3)	70.3 (7.3)	73.2 (7.4)	72.6 (7.9)	72.7 (4.1)
Difference from baseline [b]			*1.7 (2.5)*	*4.6 (6.7)*	*4.0 (5.8)*	*4.1 (6.0)**

[a] Mean (SD)
[b] Absolute difference from baseline (percentage change relative to baseline)
* Statistically significant values
CHQ-87 = Child Health Questionnaire-87, m = months, SF-36 = Short Form Survey 36, SSQ = Single Step Questionnaire, STAI-6 = Six-item State-Trait Anxiety Inventory, WHOQOL-BREF = World Health Organization Quality of Life Brief Version

SSQ

Total scores on the SSQ did change over two years (Δ4.1 (2.0-6.3)), increasing from 68.6 to 72.7 points. QoL improved until twelve months after treatment initiation, after that it remained stable, without any significant decline. During treatment scores on exercise capacity (Δ0.5 (0.2-0.7)), satisfaction with body (Δ0.3 (0.1-0.5)), impact of DCS-bracing on social life (Δ0.4 (0.2-0.6)), limitations on social activity during bracing (Δ0.5 (0.1-0.8)), self-image after start of DCS-bracing (Δ0.5 (0.2-0.8)), pain in general (Δ0.1 (0.0-0.4)), and improvement of the thorax (Δ0.2 (0.0-0.4)) increased. Scores on general health (Δ0.2 (-0.0-0.4)), limitations on social activity before DCS-bracing (Δ0.1 (-0.2-0.4)), skin problems during treatment (Δ0.1 (-0.3-0.4)), self-image before start DCS-bracing (Δ0.2 (-0.3-0.7)), pain during the application of the DCS-brace (Δ0.1 (-0.2-0.4)), pain during daily activities (Δ0.1 (-0.1-0.4)), satisfaction with the DCS-brace (Δ0.2 (-0.0-0.4)), and awareness of the DCS-brace during wearing (Δ0.3 (-0.0-0.5)) did not change. Of all patients, 96.5% (192/199) was either satisfied (42.7%, 85/199), very satisfied (34.7%, 69/199) or extremely satisfied (19.1%, 38/199) with DCS-bracing at the conclusion of their treatment or at their last assessment before dropping out. Additionally, 85.0% (170/200) of patients would opt for DCS-bracing again if they had to make the decision anew, 12.5% (25/200) had doubts and 2.5% (5/200) would definitely not choose DCS-bracing again.

Improvement in QoL and success of treatment

Overall scores on the SF-36 (Δ9.0 (3.6-14.5)), scores on the subdomains physical health (Δ9.4 (3.7-15.0)), psychological health (Δ11.9 (5.5-18.3)) and environment (Δ6.4 (0.3-12.6)) of the WHOQOL-BREF, and overall scores on the SSQ (Δ4.6 (2.4-6.7)) did change in the successfully treated group. Scores on the subdomain social relationships (Δ-0.5 (-6.7-5.7)) and scores on the CHQ-87 (Δ5.0 (-0.6-10.6)) and the STAI-6 (Δ0.5 (-0.2-1.2)) did not change. In contrast, the unsuccessfully treated group showed no improvement in the total scores of any of the questionnaires (CHQ-87 (Δ5.9 (-2.1-13.9)), SF-36 (Δ-1.5 (-15.9-12.9)), subdomains physical health (Δ-0.5 (-17.2-16.2)), psychological health (Δ6.4 (-14.3-27.2)), social relationships (Δ-2.9 (-27.7-22.0)) and environment (Δ-4.0 (-26.4-18.3)) of the WHOQOL-BREF, STAI-6 (Δ0.3 (-2.3-1.7)) and SSQ (Δ-4.7 (-11.9-2.5))). This led to overall lower scores in QoL in the unsuccessfully treated group compared to the successfully treated group. Still, 65.8% (25/38) of the unsuccessfully treated group would opt for DCS-bracing again if they had to make the decision anew and 86.5% (32/37) was either satisfied or very satisfied with the treatment at their last assessment.

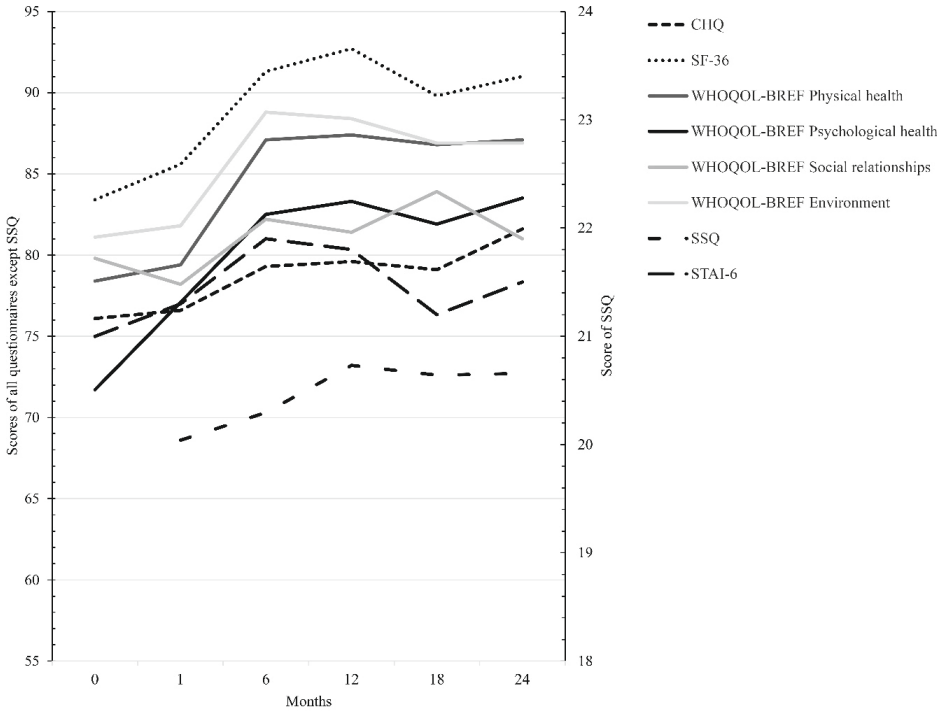


Figure 2 Change in (overall) scores of questionnaires (CHQ-87 = Child Health Questionnaire-87, SF-36 = Short Form Survey 36, SSQ = Single Step Questionnaire, STAI-6 = Six-item State-Trait Anxiety Inventory, WHOQOL-BREF = World Health Organization Quality of Life Brief Version

DISCUSSION

The overall QoL of patients treated with DCS-bracing for PC increased across the SF-36 and the WHOQOL-BREF, but not on the CHQ-87. The largest change in QoL took place in the first six to twelve months after treatment initiation, after this period QoL stabilized. There was no improvement in anxiety scores across the STAI-6. Scores on physical complains, pain, psychological health and self-esteem/self-image improved across all questionnaires. Most patients (87.2%) would choose DCS-bracing again and 94.9% were satisfied with the treatment.

In contrast to the successfully treated group, the unsuccessfully treated group showed no improvement on any of the questionnaires.

Change in QoL

The QoL measured by the CHQ-87, did not change. This may be due to the age difference between patients completing the CHQ-87 (aged 14.0 (13.0-15.0)) and those completing the SF-36 and WHOQOL-BREF (aged 17.0 (16.0-17.0)). Concerns about appearance typically begin around puberty, which tends to start later in boys. In our population, 92% of the patients are male. In a significant part of the younger group, it is likely that self-esteem concerns have not yet developed, and it is mainly the parents who are worried about the condition rather than the children themselves, often leading parents to initiate treatment decisions. In contrast, the other group consists of patients who are clearly going through puberty, resulting in increased self-awareness and self-esteem issues, prompting them to seek treatment independently. Consequently, the younger group may have a higher baseline QoL since their QoL is probably not affected, which could explain the lack of significant change in QoL observed in this group. However, assessing this difference in baseline QoL is challenging due to the use of different questionnaires across the groups.

This is one of the first studies to report on changes in QoL, making it difficult to compare our results to existing literature. The only study examining overall QoL in patients with PC reported no significant change in overall health over a follow-up period of 41 weeks.[8] Nevertheless, literature is available on related topics such as anxiety, self-esteem, and treatment satisfaction

Anxiety, physical complains, pain, psychological health and self-esteem/self-image

Earlier research suggested that PC effects patients the most on their psychological health, self-image and self-esteem but also causes physical complaints and pain.[3-6] Every questionnaire showed improvement in the physical domain, with treatment leading to improvement of physical complaints, increased exercise capacity and energy. The same applies for pain, psychological health and self-esteem/self-image. There was no improvement on state anxiety scores.

These results are consistent with the scarcely available literature. Colozza et al. reports an improvement of both chest appearance and self-esteem (+17.8%) in a small group of patients (n=20) that were successfully treated with an external brace.[7] Fraser et al. report changes on anxiety/depression (-4 on a 10-point Likert scale) and appearance/body confidence (+6 on a 10-point Likert scale) in a study with 159 patients.[8] None of the studies reported on improvement of physical complaints or pain.

Treatment evaluation

In general, patients were very satisfied with the DCS-brace, 94.9% of the patients was satisfied and 87.2% of patients were confident that they would use the brace again. This is similar to another study, which reported a satisfaction rate of 94.8%.[9] In another study the SSQ was also used, with a reported mean satisfaction of 3.7 (on a 5-point Likert-scale), comparable to our mean satisfaction of 3.8 after two years. They also reported that most patients would use the brace again.[7] Other studies used a single 5- or 10-point Likert scale to assess patients satisfaction, reporting 4.7 out of 5 and 7-8 out of 10.[8, 21]

Even when treatment fails, 65.8%% of patients would choose DCS-bracing as treatment again and 86.5% of these patients were satisfied with the treatment. This highlights the overall positive perception of DCS-bracing as a treatment for PC.

Treatment duration and results

In one of our earlier studies in the same patient population, we demonstrated improvement in self-assessment of the chest wall, with scores increasing from 4.0 to 7.8 on a 10-point Likert scale within the first six months.[6] As seen in our QoL results, improvements stabilize after six months, suggesting that most progress occurs during this initial period, which patients can notably observe. This early improvement boosts their confidence, thereby enhancing QoL scores. Although the results on improvement of QoL suggest that after six months not much progress is made, the remaining months of treatment, with a median duration of sixteen months, are crucial for maintaining the chest wall's position to prevent relapse.

Limitations

A limitation of this study is the gradual decline in response rates over time, which is anticipated as some patients complete treatment early and are subsequently lost to follow-up. Despite this, such attrition may still influence the robustness of the reported outcomes. Although the use of different questionnaires was necessary for our different aims (CHQ-87 for younger patients, SF-36 and WHOQOL-BREF for older patients, STAI-6 for anxiety, and SSQ for treatment satisfaction), questionnaire fatigue may have contributed to the decline in responses.

Additionally, the lack of a control group for comparison limits our ability to assess whether the post-treatment QoL is comparable to that of healthy individuals. Incorporating a control group in future studies would provide a stronger basis for evaluating the true impact of treatment on QoL in these patients.

Another limitation relates to not using Minimal Clinically Important Differences (MCID). These values for the WHOQOL-BREF and SF-36 have been established for chronic and severe illnesses, such as cancer, but not for pectus deformities.[22] Although some studies have cautiously applied these MCID values to less serious conditions, they typically involved populations of similar age to those in the original MCID studies and conditions more serious than pectus deformities.[23] In contrast, our study includes a significantly younger population with a much less severe condition, making the cautious application of these MCID values unsuitable in our opinion. Determining MCID values specifically for pectus deformities is necessary to accurately define what percentage increase in quality of life is clinically meaningful for this population.

Conclusions

DCS-bracing is a successful treatment in patients with PC, improving QoL in general and on different subdomains. DCS-bracing decreases physical complaints and pain and improves psychological health and self-esteem/self-image. The largest changes in QoL take place in the first six to twelve months after treatment initiation. Overall most patients are confident that they would choose DCS-bracing again and nearly all patients are satisfied with the treatment.

REFERENCES

1 Janssen N, Coorens NA, Franssen A, et al. Pectus excavatum and carinatum: a narrative review of epidemiology, etiopathogenesis, clinical features, and classification. J Thorac Dis. 2024;16(2):1687-701.

2 Van Braak H, de Beer SA, Zwaveling S, et al. Ravitch Surgery or Dynamic Compression Bracing for Pectus Carinatum: A Retrospective Cohort Study. Ann Thorac Surg. 2024;117(1):144-50.

3 Krille S, Müller A, Steinmann C, et al. Self- and social perception of physical appearance in chest wall deformity. Body Image. 2012;9(2):246-52.

4 Steinmann C, Krille S, Mueller A, et al. 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. Eur J Cardiothorac Surg. 2011;40(5):1138-45.

5 Koumbourlis AC. Pectus deformities and their impact on pulmonary physiology. Paediatr Respir Rev. 2015;16(1):18-24.

6 De Beer S, Volcklandt S, de Jong J, et al. Dynamic Compression Therapy for Pectus Carinatum in Children and Adolescents: Factors for Success. J Pediatr Surg. 2023;58(8):1440-5.

7 Colozza S, Bütter A. Bracing in pediatric patients with pectus carinatum is effective and improves quality of life. J Pediatr Surg. 2013;48(5):1055-9.

8 Fraser S, Harling L, Patel A, Richards T, Hunt I. External Compressive Bracing With Initial Reduction of Pectus Carinatum: Compliance Is the Key. Ann Thorac Surg. 2020;109(2):413-9.

9 Ermerak NO. Compressive external bracing in pectus carinatum : Results of the first 100 patients. Marmara Med J. 2023;36(3):334-8.

10 Knudsen MV, Grosen K, Pilegaard HK, Laustsen S. Surgical correction of pectus carinatum improves perceived body image, mental health and self-esteem. J Pediatr Surg. 2015;50(9):1472-6.

11 Bostanci K, Ozalper MH, Eldem B, et al. Quality of life of patients who have undergone the minimally invasive repair of pectus carinatum. Eur J Cardiothorac Surg. 2013;43(1):122-6.

12 Krasopoulos G, Dusmet M, Ladas G, Goldstraw P. Nuss procedure improves the quality of life in young male adults with pectus excavatum deformity. Eur J Cardiothorac Surg. 2006;29(1):1-5.

13 Marteau TM, Bekker H. The development of a six-item short-form of the state scale of the Spielberger State-Trait Anxiety Inventory (STAI). Br J Clin Psychol. 1992;31(3):301-6.

14 Landgraf J, Abetz L, Ware J. The CHQ User's Manual. Boston, The Health Institute, New England Medical Center; 1996.

15 Skevington SM, Lotfy M, O'Connell KA. The World Health Organization's WHOQOL-BREF quality of life assessment: psychometric properties and results of the international field trial. A report from the WHOQOL group. Qual Life Res. 2004;13(2):299-310.

16 Aaronson NK, Muller M, Cohen PD, et al. Translation, validation, and norming of the Dutch language version of the SF-36 Health Survey in community and chronic disease populations. J Clin Epidemiol. 1998;51(11):1055-68.

17 Farivar SS, Cunningham WE, Hays RD. Correlated physical and mental health summary scores for the SF-36 and SF-12 Health Survey, V.I. Health Qual Life Outcomes. 2007;5:54.

18 Lins L, Carvalho FM. SF-36 total score as a single measure of health-related quality of life: Scoping review. SAGE Open Med. 2016;4:2050312116671725.

19 Twisk JWR. Applied Longitudinal Data Analysis for Epidemiology: A Practical Guide. 2 ed. Cambridge: Cambridge University Press; 2013.

20 Kravarusic D, Dicken BJ, Dewar R, et al. The Calgary protocol for bracing of pectus carinatum: a preliminary report. J Pediatr Surg. 2006;41(5):923-6.

21 Martinez-Ferro M, Fraire C, Bernard S. Dynamic compression system for the correction of pectus carinatum. Semin Pediatr Surg. 2008;17(3):194-200.

22 Mouelhi Y, Jouve E, Castelli C, Gentile S. How is the minimal clinically important difference established in health-related quality of life instruments? Review of anchors and methods. Health Qual Life Outcomes. 2020;18(1):136.

23 Vranceanu AM, Manglani HR, Choukas NR, et al. Effect of Mind-Body Skills Training on Quality of Life for Geographically Diverse Adults With Neurofibromatosis: A Fully Remote Randomized Clinical Trial. JAMA Netw Open. 2023;6(6):e2320599.

APPENDIX A

SSQ

Circle the number that best matches how you felt about the question in the past week. For questions 8 and 9, you may write down a number, where 0 represents the worst self-image you can imagine and 10 represents the best self-image.

1	General health **after** brace therapy	5 much better now 4 slightly better 3 about the same 2 slightly worse now 1 much worse now
2	Exercise capacity **after** brace therapy	5 much better now 4 slightly better 3 about the same 2 slightly worse now 1 much worse now
3	Extent to which social activity is hindered by the appearance of the chest **before** starting brace therapy	5 extremely 4 quite a lot 3 moderately 2 a little 1 not at all
4	Extent to which social activity is hindered by the appearance of the chest **after** brace therapy	5 not at all 4 quite a lot 3 moderately 2 a little 1 extremely
5	Satisfaction with overall appearance **after** brace therapy	5 extremely satisfied 4 very satisfied 3 satisfied 2 dissatisfied 1 very dissatisfied
6	Problems with the skin under the pad	5 not at all 4 very little 3 little 2 a bit 1 a lot
7	Impact of the brace on social life	5 much better now 4 slightly better 3 about the same 2 slightly worse now 1 much worse now
8	Self-esteem **before** brace treatment	score 0-10
9	Self-esteem during or **after** brace treatment	score 0-10

10	Pain during the start of brace therapy	5 none 4 very little 3 little 2 a lot 1 very much
11	Pain interfering with day-to-day activity now	5 not at all 4 very little 3 little 2 a bit 1 a lot
12	Pain now	5 none 4 occasionally 3 moderate, no pain medication 2 moderate, with pain medication 1 a lot
13	Awareness of the brace while wearing it	5 not at all 4 little 3 moderate 2 a lot 1 extreme
14	Overall satisfaction with the final result	5 extremely satisfied 4 very satisfied 3 satisfied 2 dissatisfied 1 very dissatisfied
15	Chest looks different	5 much better now 4 slightly better 3 about the same 2 slightly worse now 1 much worse now
16	Going back, would have started the brace therapy again	10 yes 5 doubtful 0 no

APPENDIX B

STAI-state

Below you will find a number of statements that people have used to describe themselves. Read each statement and then circle the number on the right of the statement that best indicates how you feel right now, at this moment. There are no right or wrong answers. Don't think too long, and go with your first impression, as it is usually the best. The goal is to reflect how you feel at this moment.

		1 = NOT AT ALL	2 = A LITTLE	3 = QUITE A BIT	4 = VERY MUCH
1.	I feel calm	1	2	3	4
2.	I am tense	1	2	3	4
3.	I am confused	1	2	3	4
4.	I am relaxed	1	2	3	4
5.	I feel content	1	2	3	4
6.	I am worried	1	2	3	4

APPENDIX C

CHQ-87

Part 1: Your Health				
1.1 How would you describe your health?				
1 ❑ excellent	2 ❑ very good	3 ❑ good	4 ❑ fair	5 ❑poor

Part 2: Your Physical Activities					
The following questions are about physical activities you can do on a normal day.					
2.1 In the **past 4 weeks**, has it been difficult for you to do the following activities because of your health?					
(Complete all questions)		Yes, very difficult	Yes, difficult	Yes, somewhat difficult	No, not difficult
		1	2	3	4
a	things that require a lot of effort, like playing football or running	❑	❑	❑	❑
b	things that require some effort, like cycling or (roller) skating	❑	❑	❑	❑
c	walking a long distance or climbing several flights of stairs	❑	❑	❑	❑
d	walking around school or the neighborhood	❑	❑	❑	❑
e	walking a short distance or climbing one flight of stairs	❑	❑	❑	❑
f	doing your household chores	❑	❑	❑	❑
g	bending, lifting, or stooping	❑	❑	❑	❑
h	eating, dressing, washing, or going to the toilet by yourself	❑	❑	❑	❑
i	getting in and out of bed or getting up from a chair	❑	❑	❑	❑

Part 3: Your Daily Activities

3.1 In the **past 4 weeks**, has it been difficult for you, due to feeling sad or worried, to:

(Complete all questions)		Yes, very difficult	Yes, difficult	Yes, somewhat difficult	No, not difficult
		1	2	3	4
a	do certain types of schoolwork or things with friends?	❑	❑	❑	❑
b	spend the normal amount of time on your schoolwork and friends?	❑	❑	❑	❑
c	finish your schoolwork or do anything with your friends?	❑	❑	❑	❑

3.2 In the **past 4 weeks**, due to problems with your behavior, has it been difficult for you to:

(Complete all questions)		Yes, very difficult	Yes, difficult	Yes, somewhat difficult	No, not difficult
		1	2	3	4
a	do certain types of schoolwork or things with friends?	❑	❑	❑	❑
b	spend the normal amount of time on your schoolwork and friends?	❑	❑	❑	❑
c	finish your schoolwork or do anything with your friends?	❑	❑	❑	❑

3.3 In the **past 4 weeks**, due to problems with your physical health, has it been difficult for you to:

(Complete all questions)		Yes, very difficult	Yes, difficult	Yes, somewhat difficult	No, not difficult
		1	2	3	4
a	do certain types of schoolwork or things with friends?	❑	❑	❑	❑
b	spend the normal amount of time on your schoolwork and friends?	❑	❑	❑	❑
c	finish your schoolwork or do anything with your friends?	❑	❑	❑	❑

	Part 4: Pain
4.1 How much physical pain or discomfort have you had in the **past 4 weeks**?	
1	❑ none
2	❑ a little bit
3	❑ a bit
4	❑ quite a lot
5	❑ a lot
6	❑ very much

4.2 How often have you experienced this physical pain or discomfort in the **past 4 weeks**?	
1	❑ never
2	❑ once or twice
3	❑ a few times
4	❑ often
5	❑ very often
6	❑ (almost) every day

Part 5: Behavior						
Below is a list of ways young people behave and the problems they sometimes have.						
5.1 How often in the **past 4 weeks**:						
(Complete all questions)		Very often	Often	Sometimes	Almost never	Never
		1	2	3	4	5
a	did you behave childishly?	❑	❑	❑	❑	❑
b	did you argue?	❑	❑	❑	❑	❑
c	did you find it difficult to concentrate on something?	❑	❑	❑	❑	❑
d	did you not do what your teacher or parents asked you to do?	❑	❑	❑	❑	❑
e	did you want to be alone?	❑	❑	❑	❑	❑
f	did you lie or deceive someone?	❑	❑	❑	❑	❑
g	did you struggle to be liked?	❑	❑	❑	❑	❑
h	did you feel awkward?	❑	❑	❑	❑	❑
i	did you run away from home?	❑	❑	❑	❑	❑
j	did you have speech problems, such as stuttering?	❑	❑	❑	❑	❑
k	did you steal something at home?	❑	❑	❑	❑	❑
l	did you steal something outside home?	❑	❑	❑	❑	❑
m	did you become cranky or moody when you didn't get your way?	❑	❑	❑	❑	❑
n	did you get very angry when you didn't get your way?	❑	❑	❑	❑	❑
o	did you find it difficult to do things with others?	❑	❑	❑	❑	❑
p	did you find it difficult to get along with others?	❑	❑	❑	❑	❑

5.2 How would you generally describe your behavior compared to other children your age?				
1 ❑ excellent	2 ❑ very good	3 ❑ good	4 ❑ fair	5 ❑ poor

Part 6: Your Feelings						
The following sentences are about moods and feelings that young people can have.						
6.1 How often in the **past 4 weeks:**						
(Complete all questions)		Always	Usually	Sometimes	Almost never	Never
		1	2	3	4	5
a	did you feel sad?	❑	❑	❑	❑	❑
b	did you feel like crying?	❑	❑	❑	❑	❑
c	did you feel anxious or scared?	❑	❑	❑	❑	❑
d	did you worry about something?	❑	❑	❑	❑	❑
e	did you feel lonely?	❑	❑	❑	❑	❑
f	did you feel unhappy?	❑	❑	❑	❑	❑
g	did you feel nervous?	❑	❑	❑	❑	❑
h	did you feel upset?	❑	❑	❑	❑	❑
i	did you feel happy?	❑	❑	❑	❑	❑
j	did you feel cheerful?	❑	❑	❑	❑	❑
k	did you enjoy the things you did?	❑	❑	❑	❑	❑
l	did you have fun?	❑	❑	❑	❑	❑
m	did you feel restless?	❑	❑	❑	❑	❑
n	did you have trouble sleeping?	❑	❑	❑	❑	❑
o	did you have a headache?	❑	❑	❑	❑	❑
p	were you satisfied with yourself?	❑	❑	❑	❑	❑

Part 7: Self-Confidence						
What do you think of yourself, your school, and others? It may help to think about what other children your age would think of these things.						
7.1 How satisfied or dissatisfied have you been in the **past 4 weeks** with:						
(Complete all questions)		Very satisfied	Satisfied	Neither satisfied nor dissatisfied	Dissatisfied	Very dissatisfied
		1	2	3	4	5
a	yourself	❑	❑	❑	❑	❑
b	your schoolwork	❑	❑	❑	❑	❑
c	how good you are at sports	❑	❑	❑	❑	❑
d	your friendships	❑	❑	❑	❑	❑
e	the things you are capable of	❑	❑	❑	❑	❑
f	how well you get along with others	❑	❑	❑	❑	❑
g	your body and appearance	❑	❑	❑	❑	❑
h	how you usually feel	❑	❑	❑	❑	❑
i	how well you get along with your family members	❑	❑	❑	❑	❑
j	how life is for you	❑	❑	❑	❑	❑
k	how good you are at being a friend to others	❑	❑	❑	❑	❑
l	what others think of you	❑	❑	❑	❑	❑
m	how you can talk with others	❑	❑	❑	❑	❑
n	your overall health	❑	❑	❑	❑	❑

Part 8: Your Health						
Below are statements about health in general.						
8.1 How true or not true is the statement for you?						
(Complete all questions)		True	Usually true	Don't know	Usually not true	Not true
		1	2	3	4	5
a	my health is excellent	❑	❑	❑	❑	❑
b	I have been so sick once that I thought I would die	❑	❑	❑	❑	❑
c	I am never very sick	❑	❑	❑	❑	❑
d	I am less healthy than other young people	❑	❑	❑	❑	❑
e	I have never been very sick	❑	❑	❑	❑	❑
f	I seem to always get sick	❑	❑	❑	❑	❑
g	I think I will be less healthy later on	❑	❑	❑	❑	❑
h	I think I will be very healthy later on	❑	❑	❑	❑	❑
i	I never worry about my health	❑	❑	❑	❑	❑
j	I think I am healthy now	❑	❑	❑	❑	❑
k	I worry more about my health than my peers	❑	❑	❑	❑	❑

8.2 How would you describe your health now compared to one year ago?	
1	❑ now much better than one year ago
2	❑ now slightly better than one year ago
3	❑ now about the same as one year ago
4	❑ now slightly worse than one year ago
5	❑ now much worse than one year ago

Part 9: You and Your Family						
9.1 How often, in the **past 4 weeks**, has your health or behavior:						
(Complete all questions)		Very often	Often	Sometimes	Almost never	Never
		1	2	3	4	5
a	limited the types of activities your family could do?	❑	❑	❑	❑	❑
b	disrupted everyday family activities (like eating or watching TV)?	❑	❑	❑	❑	❑
c	contributed to your family not being able to go out unexpectedly?	❑	❑	❑	❑	❑
d	led to tensions or conflicts at home?	❑	❑	❑	❑	❑
e	been a source of disagreement or bickering in your family?	❑	❑	❑	❑	❑
f	caused your family to cancel or change plans at the last moment?	❑	❑	❑	❑	❑

9.2 Sometimes family members don't get along very well. They don't always agree and can get angry. How well do you think your family gets along at home?				
1 ❑ excellent	2 ❑ very good	3 ❑ good	4 ❑ fair	5 ❑ poor

APPENDIX D

WHOQOL-Bref

We ask you to indicate in this questionnaire what you think of your quality of life, health, and other life areas. **Please answer all the questions.** If you are unsure about how to answer a question, **choose the response that seems most applicable**, often your first reaction. Keep your standards, hopes, pleasures, and concerns in mind. Think about your life in the **last two weeks**. For example, a question might be:

	Not at all	A little	Moderately	A lot	An extreme amount
Do you receive the kind of support you need from others?	1	2	3	4	5

You should circle the number that best matches the amount of support you have received from others in the past two weeks. For example, you would circle the number 4 if you received a lot of support from others, as shown below:

	Not at all	A little	Moderately	A lot	An extreme amount
Do you receive the kind of support you need from others?	1	2	3	4	5

Now the actual questionnaire begins:

		Very poor	Poor	Neither poor nor good	Good	Very good
1(G1)	How would you rate your quality of life?	1	2	3	4	5

		Very dissatisfied	Dissatisfied	Neither satisfied nor dissatisfied	Satisfied	Very satisfied
2(G4)	How satisfied are you with your health?	1	2	3	4	5

The following questions ask **to what extent (how much)** you have experienced certain things in the past two weeks.

		Not at all	A little	Moderately	A lot	An extreme amount
3(F1.2)	Do you worry about your pain or discomfort?	1	2	3	4	5
4(F1.3)	How difficult is it for you to deal with pain or discomfort?	1	2	3	4	5
5(F1.4)	To what extent do you feel that pain prevents you from doing what you need to do?	1	2	3	4	5
6(F11.3)	How much do you need medical treatment to function in your daily life?	1	2	3	4	5
7(F4.1)	How much do you enjoy life?	1	2	3	4	5
8(F24.2)	To what extent do you feel your life is meaningful?	1	2	3	4	5

The following questions ask **to what extent** you have experienced certain things or were able to do certain things in the past two weeks.

		Not at all	A little	Moderately	A lot	An extreme amount
9(F5.3)	How well are you able to concentrate?	1	2	3	4	5
10(F7.2)	Do you feel hindered by your appearance?	1	2	3	4	5
11(F7.3)	Is there anything about your appearance that makes you feel uncomfortable?	1	2	3	4	5
12(F16.1)	How safe do you feel in your daily life?	1	2	3	4	5
13(F22.1)	How healthy is your environment?	1	2	3	4	5
14(F2.1)	Do you have enough energy for daily life?	1	2	3	4	5
15(F7.1)	Can you accept your physical appearance?	1	2	3	4	5

16(F18.1)	Do you have enough money to meet your needs?	1	2	3	4	5
17(F20.1)	How available to you is the information you need in your daily life?	1	2	3	4	5
18(F21.1)	Do you have opportunities for leisure activities?	1	2	3	4	5

The following questions ask how **satisfied or dissatisfied** you have been with various aspects of your life in the past two weeks.

		Very dissatisfied	**Dissatisfied**	**Neither satisfied nor dissatisfied**	**Satisfied**	**Very satisfied**
19(F3.3)	How satisfied are you with your sleep?	1	2	3	4	5
20(F7.4)	How satisfied are you with your appearance?	1	2	3	4	5
21(F10.3)	Are you satisfied with your ability to perform daily activities?	1	2	3	4	5
22(F12.4)	Are you satisfied with your work capacity?	1	2	3	4	5
23(F6.3)	Are you satisfied with yourself?	1	2	3	4	5
24(F13.3)	How satisfied are you with your personal relationships?	1	2	3	4	5
25(F15.3)	To what extent are you satisfied with your sex life?	1	2	3	4	5
26(F14.4)	How satisfied are you with the support you get from friends?	1	2	3	4	5
27(F17.3)	Are you satisfied with your living conditions?	1	2	3	4	5
28(F19.3)	How satisfied are you with your access to health services?	1	2	3	4	5
29(F23.3)	How satisfied are you with your transport?	1	2	3	4	5

		Very poor	Poor	Neither poor nor good	Good	Very good
30(F9.1)	How well are you able to move around?	1	2	3	4	5

The following question refers to **how often** you have felt or experienced certain things, such as support from your family or friends, or negative experiences, like feeling unsafe.

		Never	Rarely	Sometimes	Fairly often	Always
31(F1.1)	How often have you experienced pain?	1	2	3	4	5
32 (F8.1)	How often have you experienced negative feelings such as depression or anxiety?	1	2	3	4	5

APPENDIX E

SF-36

This questionnaire is about your views on your health. These data can help track how you feel and how well you can perform your usual activities. Answer each question by marking the response as indicated. If you are unsure about how to answer a question, give the best possible answer.

1. How would you rate your general health? (circle one number)	
Excellent	1
Very good	2
Good	3
Fair	4
Poor	5

2. How would you rate your health now compared to a year ago? (circle one number)	
Much better now than a year ago	1
Somewhat better now than a year ago	2
About the same as a year ago	3
Somewhat worse now than a year ago	4
Much worse now than a year ago	5

3. The following questions are about activities you might do on a typical day. Are you limited in these activities because of your health? If so, how much?

	Activities	**Yes, severely limited**	**Yes, slightly limited**	**No, not limited at all**
a	**Vigorous activities** such as running, lifting heavy objects, participating in strenuous sports	1	2	3
b	**Moderate activities** such as moving a table, vacuuming, swimming, or cycling	1	2	3
c	Lifting or carrying groceries	1	2	3
d	Climbing **several** flights of stairs	1	2	3
e	Climbing **one** flight of stairs	1	2	3
f	Bending, kneeling, or squatting	1	2	3
g	Walking **more than a kilometer**	1	2	3
h	Walking **several hundred meters**	1	2	3
i	Walking about **one hundred meters**	1	2	3
j	Bathing or dressing yourself	1	2	3

4.	**In the past 4 weeks, have you had any of the following problems at work or with other daily activities due to your physical health? (circle one number)**	**Yes**	**No**
a	Spent **less time** on work or other activities	1	2
b	Accomplished **less** than you would like	1	2
c	Limited in the **kind of work** or other activities	1	2
d	Had **difficulty** performing work or other activities (for example, it took extra effort)	1	2

5.	**In the past 4 weeks, have you had any of the following problems with your work or other daily activities due to emotional problems (such as feeling depressed or anxious)? (circle one number)**	**Yes**	**No**
a	Spent **less time** on work or other activities	1	2
b	Accomplished **less** than you would like	1	2
c	Didn't do work or other activities **as carefully** as usual	1	2

6. **To what extent have your physical health or emotional problems interfered with your normal social activities with family, friends, neighbors, or groups during the past 4 weeks? (circle one number)**	
Not at all	1
Slightly	2
Moderately	3
Quite a bit	4
Extremely	5

7. **How much bodily pain have you had during the past 4 weeks? (circle one number)**	
None	1
Very mild	2
Mild	3
Moderate	4
Severe	5
Very severe	6

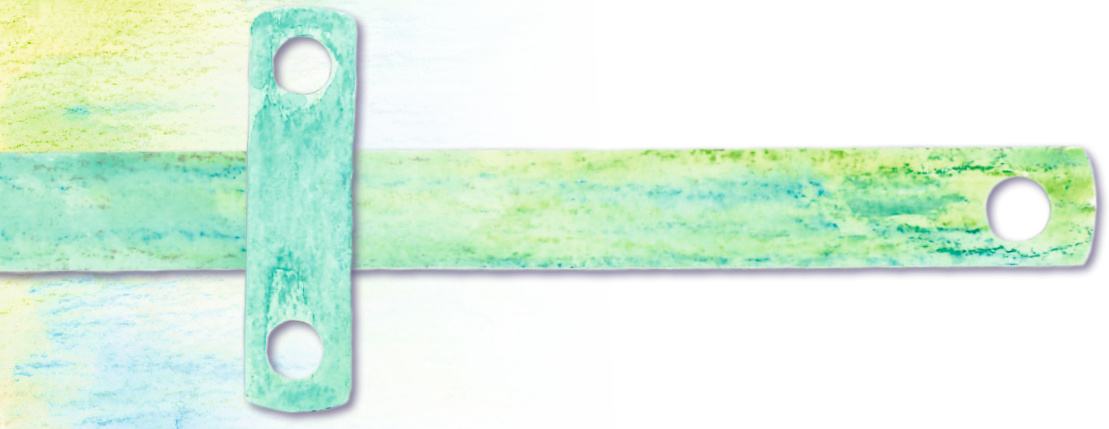
8. During the past 4 weeks, how much did pain interfere with your normal work (including work outside the home and housework)? (circle one number)	
Not at all	1
Slightly	2
Moderately	3
Quite a bit	4
Extremely	5

9. These questions are about how you feel and how things have been with you during the past 4 weeks. Please give the answer that comes closest to how you have felt. (circle one number)

		Always	Usually	Often	Sometimes	Rarely	Never
a	Did you feel full of life?	1	2	3	4	5	6
b	Were you very nervous?	1	2	3	4	5	6
c	Did you feel so down that nothing could cheer you up?	1	2	3	4	5	6
d	Did you feel calm and content?	1	2	3	4	5	6
e	Did you have a lot of energy?	1	2	3	4	5	6
f	Did you feel gloomy and downhearted?	1	2	3	4	5	6
g	Did you feel exhausted?	1	2	3	4	5	6
h	Were you a happy person?	1	2	3	4	5	6
i	Did you feel tired?	1	2	3	4	5	6

10. How often during the past 4 weeks did your physical health or emotional problems interfere with your social activities (like visiting friends or relatives)? (circle one number)	
Always	1
Usually	2
Sometimes	3
Rarely	4
Never	5

11.	How TRUE or FALSE is <u>each</u> of the following statements for you? (circle one number)					
		Absolutely true	**Mostly true**	**Don't know**	**Mostly false**	**Absolutely false**
a	I seem to get sick a little easier than other people	1	2	3	4	5
b	I am as healthy as anybody I know	1	2	3	4	5
c	I expect my health to get worse	1	2	3	4	5
d	My health is excellent	1	2	3	4	5



Chapter 4

Evaluating inter- and intraobserver agreement on pectus carinatum severity and treatment outcomes: a comparison of subjective and objective assessment methods

Hendrik van Braak, Sjoerd A. de Beer, Sander Zwaveling, Matthijs W.N. Oomen, L.W. Ernest van Heurn, Justin R. de Jong

European Journal of Pediatric Surgery. 2025 June 35:232-239.

ABSTRACT

Background

Visual examination is crucial for assessing pectus carinatum (PC) severity and treatment results. This cross-sectional study evaluates the inter- and intraobserver agreement of PC deformities before and after treatment.

Methods

Observers examined medical photographs of patients before and after treatment. Primary outcome was inter- and intraobserver agreement on esthetic results after treatment. Secondary outcomes included inter- and intraobserver agreement on severity and symmetry before treatment, differences in esthetic results after Ravitch surgery and dynamic compression bracing (DCS-bracing), and the impact of scars, age and treatment duration on esthetic results.

Results

Medical photographs of 201 patients (aged 4-18) were evaluated by five surgeons and five peers. Surgeons and peers demonstrated inadequate (κ<.61) interobserver agreement on esthetic results (κ=.26, κ=.22), severity of PC (κ=.43, κ=.38) and symmetry (κ=.37, surgeons only). Agreement between surgeons and peers on esthetic results (κ=.37) and severity before treatment (κ=.54) was similarly inadequate. Surgeons and peers demonstrated inadequate intraobserver agreement on esthetic results (κ=.49, κ=.34), severity of PC (κ=.54, κ=.48) and symmetry (κ=.60, surgeons only). Deformities treated with Ravitch surgery were perceived as more severe but yielded better results. Peers, unlike surgeons, viewed scars as negatively impacting results. No relationship was found between results after treatment and treatment duration (P=.682, P=.062) or age (P=.205, P=.527).

Conclusions

Subjective assessment of PC severity and esthetic results is inconsistent. 3D scanning could help standardize treatment completion and aid patients and surgeons in determining treatment completion. The psychosocial effects of scars should be addressed when discussing treatment options.

INTRODUCTION

Pectus carinatum (PC) is a common pediatric condition characterized by overgrowth of costal cartilage which causes protrusion of the sternum and adjacent cartilage. In our institution, patients with PC were previously treated with surgical options like the Ravitch procedure and Abramson procedures. However, non-invasive treatment using Dynamic Compression System (DCS) bracing has become the preferred treatment.[1-4] Although treatment outcomes are similar,[5] a significant clinical challenge persists. Surgical outcomes are typically definitive, providing clear correction, but determining endpoints for DCS-bracing therapy remains challenging.

The subjective nature of PC's esthetic impact makes determining treatment success challenging. While objective tools like 3D scanning and the Haller index provide measurable data on the reduction of the chest deformity, [6, 7] they do not fully capture the patient's own view of success. At our institution, patients expectations vary widely; some are satisfied with modest improvement, while others aim for an ideal chest appearance. As a result, even with objective measurements, outcomes ultimately hinge on the patients' own view of success and satisfaction, in mutual agreement with the physician.[5, 8, 9] Differences in treatment success expectations between patients and physicians can influence treatment decisions, like whether or not to start the retainer phase, and treatment duration, raising questions about consistency in evaluating treatment results. We aimed to assess whether these inconsistencies exist.

The aim of this study was to evaluate the inter- and intraobserver agreement, between surgeons and peers, primarily on esthetic results after treatment and secondary on severity and symmetry before treatment. Additionally we assessed differences in average ratings between surgeons and peers and explored the influence of scars, age and treatment duration on esthetic results.

MATERIALS AND METHODS

Study design

At time of study conceptualization, we already had a sufficient collection of medical photographs available for analysis. This allowed us to conduct a retrospective cross-sectional study wherein medical photographs of pediatric patients with pectus carinatum were visually evaluated by reviewers. A METC official waiver of ethical approval was granted from the METC of the Amsterdam Medical Center. The need for informed consent was waived.

Patients and reviewers

All patients who successfully finished treatment for pectus carinatum with DCS-bracing or Ravitch surgery in the Amsterdam Pectus Center between 2009 and 2019, and had before and after treatment photographs taken, were included. Treatment with DCS-bracing was regarded successful if patients achieved a satisfactory result, in consultation with the clinician. The results of Ravitch surgery were consistently regarded as successful, as the procedure allows for a precise and complete correction of the deformity. Figure 1 shows the diagnostic and therapeutic flowchart used in treatment. Eight medical photographs (four before and four after treatment (Figure 2 and Figure 3) were taken from different angles as part of our standard protocol. Photographs were intended to be taken from every patient, not just the extremes.

Photographs were reviewed by five pediatric surgeons and five peers of patients. The limited sample size reflects the exploratory nature of this study in assessing observer agreement on esthetic outcomes.

Peers

We refer to individuals who share similar age-related characteristics with the patients as peers. Peers were selected instead of patients to provide an external perspective on esthetic outcomes, one that is less influenced by personal treatment experiences or individual biases. These peers were not directly involved in the patients' care nor were they family members and were recruited by posting an open invitation at a local high school and university, asking students to participate in the research. The peers, aged 14, 14, 16, 16 and 23 years, included three males and two females. Their age matches the age of patients, to offer a perspective more aligned with the patients' age related social and esthetic standards, which can be particularly sensitive in adolescent and young adult populations. The number of peers was matched with the number of surgeons to ensure a balanced evaluation, minimizing potential biases from unequal sample sizes.

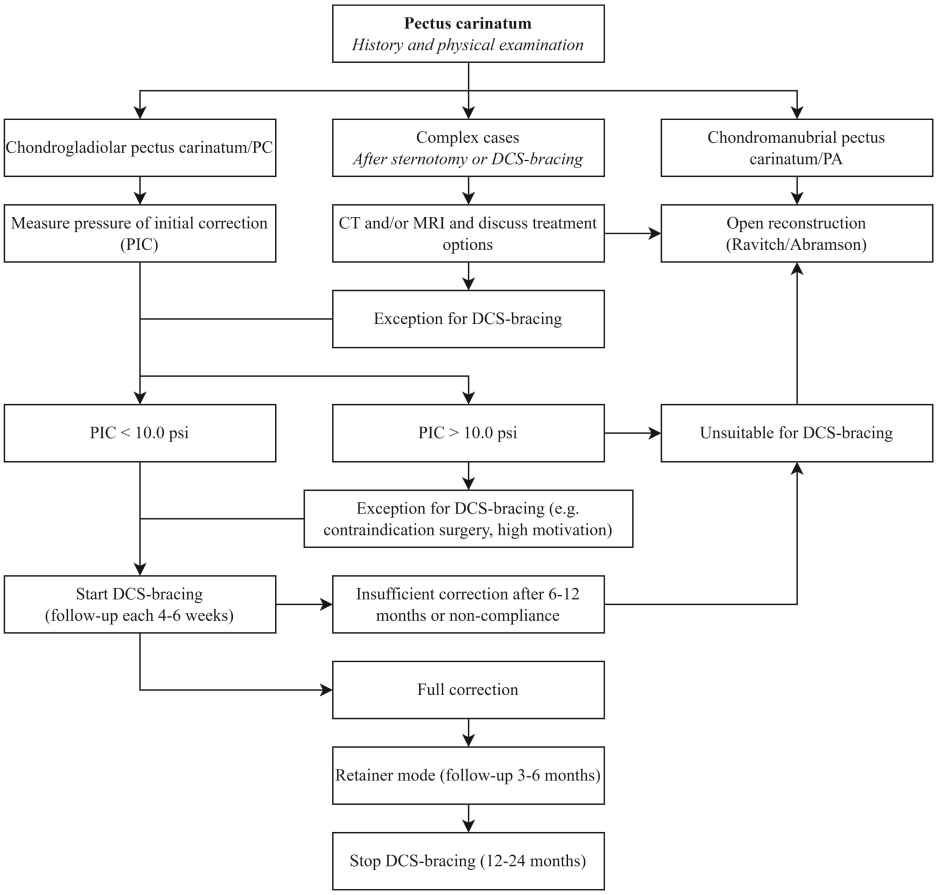


Figure 1 Diagnostic and therapeutic flowchart (CT = Computed Tomography, MRI = Magnetic Resonance Imaging, PIC = Pressure of Initial Correction, psi = pounds per square inch)

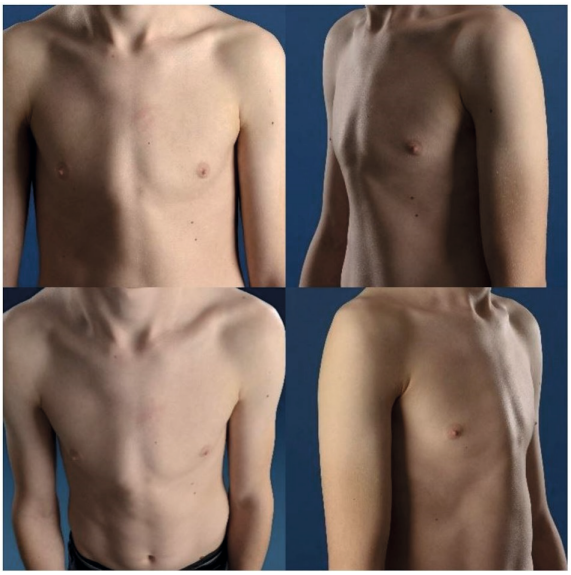


Figure 2 Example of four photographs of a patient before treatment

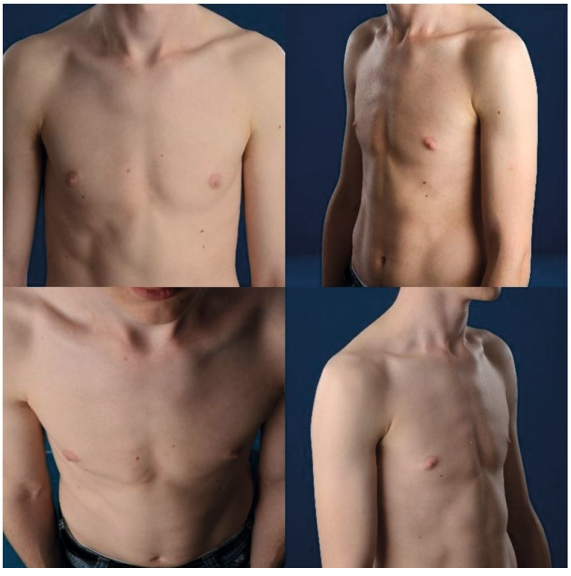


Figure 3 Example of four photographs of a patient after treatment

Data collection and measurements

Data were obtained retrospectively from patient records. Observers were asked to visually examine medical photographs of PC patients and rate the severity before treatment on a three point scale (one = mild, two = moderate, three = severe) and judge the results after treatment (one = minimal improvement, two = moderate improvement, three = good improvement). Both surgeons and peers were asked to assess esthetic results of patients who underwent surgery with the scar present, and subsequently, visualize the photographs without the scar, providing an extra rating. The scars were not digitally removed, patients were just asked to imagine the photograph without the scar. Furthermore surgeons were asked to determine if the deformity before treatment was symmetrical or not. This additional question was included exclusively for surgeons, as this factor can influence treatment selection and approach. This question was not posed to peers because they lack the clinical context to understand how symmetry affects treatment decisions, rendering the question redundant. Observers were blinded to all clinical information and completed the assessment twice, with at least a two weeks interval between both assessments.

Outcomes

Primary outcomes were interobserver and intraobserver agreement on esthetic results after treatment, secondary outcomes were inter- and intraobserver agreement on severity and symmetry before treatment, differences in average ratings between the Ravitch and DCS-bracing group and the influence of scars, age and treatment duration on esthetic results after treatment.

Statistical analysis

Descriptive measurements were utilized to characterize the study population. To assess interobserver agreement on esthetic results, severity and symmetry, we applied Cohen's kappa statistics to all pairs of observers using ratings from the initial assessment round. Cohen's kappa was selected because it accounts for chance agreement, providing a more robust measure of agreement than simple percent agreement. The weighted arithmetic mean of all observer pairs, determined by the number of evaluated subjects, was adopted as a measure of overall agreement.[10] Additionally, the interobserver agreement per observer was based on the weighted arithmetic mean of all observer pairs that included the observer in question. We computed two-sided 95% confidence intervals (95% CI). A similar approach was taken for intraobserver and scar-versus-no-scar agreement, but this time, we involved repeated assessments by the same surgeon. Average agreement was based on the mean of all observers. Prevalence-Adjusted Bias-Adjusted Kappa (PABAK) was employed to assess agreement when there was substantial disparity in the prevalence of response options.[11] PABAK was specifically chosen as it

adjusts for imbalances in response distribution, allowing a clearer understanding of agreement where one response may be more common than others.

The interpretation of kappa coefficients was as follows: coefficients ranging from .00 to .20, .21 to .40, .41 to .60, .61 to .80, and .81 to 1.00 corresponded respectively to slight, fair, moderate, substantial, and almost perfect agreement. These ranges provide context for the clinical significance of observed agreement, with higher values indicating more reliable consistency among observers, a kappa value of .61 or above is typically considered adequate agreement, indicating a substantial level of consistency beyond chance.[12]

We used Spearman's rank correlation to evaluate the statistical relationship between average ratings on severity before treatment and improvement after treatment. Descriptive statistics were used to calculate differences in average ratings. Descriptive statistics were used to analyze the relationship between treatment duration and age on results after treatment. Statistical significance was determined at a probability value of less than 0.05. Data were analyzed using IBM SPSS Statistics 28.0.

RESULTS

Between September 2009 and December 2019 a total of 736 patients underwent either Ravitch surgery or DCS-bracing for PC in the Amsterdam Pectus Center. Among these, 68.6% (505/736) completed treatment successfully. Medical photographs were taken before and after treatment of 201 patients (aged 4-18) out of this group (39.8%, 201/505). While protocol dictates photographing all pectus carinatum patients who finalize treatment, not all attended their medical photography appointments. Both pre- and posttreatment medical photographs are needed to be eligible for evaluation. The majority of included patients were male (96.5%, 194/201), with an median age of 15.0 (IQR 13.5-16.0) years. DCS-bracing was the primary treatment for most patients (69.2%, 139/201), while the remaining patients underwent Ravitch surgery (30.8%, 62/201). Six (9.7%) patients who underwent Ravitch surgery had previously failed DCS-bracing treatment. Median age in the Ravitch group was 15.0 (IQR 14.0-16.0) years compared to 14.0 (IQR 13.0-15.0) years in the DCS-bracing group (P<.001).

Interobserver agreement on severity before treatment, symmetry and results after treatment

Table 1 shows the results of the interobserver agreement. Overall, agreement was inadequate across all assessments, as all kappa values were below .61, indicating less than substantial consistency.

Fair agreement was reached among both surgeons and peers (κ=.26(.21-.31) and κ=.22(.19-.26) resp.) on results after treatment. The overall agreement of severity before treatment among surgeons showed a moderate agreement (κ=.43(.37-.49)). This was slightly higher than the fair agreement observed among peers (κ=.38(.34-.41)). Surgeons exhibited fair agreement on symmetry (κ=.37(.29-.45)). There was a moderate agreement between surgeons and peers on severity before treatment (κ=.54(.48-.60)) and a fair agreement on results after treatment (κ=.37(.30-.45)).

There was a significant relationship between average rating on severity before treatment and improvement after treatment (surgeons and peers r=.39, P<.001, r=.43, P<.001 resp.), with more severe deformities resulting in more noticeable improvements.

Intraobserver agreement on severity before treatment, symmetry and results after treatment

Table 2 shows the results of the intraobserver agreement. Overall, agreement was inadequate across all assessments, as all kappa values were below 0.61, indicating less than substantial consistency.

Surgeons exhibited more consistent results on severity before treatment (κ=.54(.47-.61)) and results after treatment (κ=.49(.40-.58)) compared to peers (κ=.48(.38-.59) and κ=.34(.28-.40) resp.). Surgeons showed an moderate average intraobserver agreement (κ=.60(.46-.74)) on symmetry.

Table 1. Interobserver agreement between surgeons and peers

	Severity before treatment		**Results after treatment** [a]
	Severity [a]	*Symmetry* [a]	
Surgeons			
Overall agreement	.43 (.37-.49)	.37 (.29-.45)	.26 (.21-.31)
Surgeon A versus B-E	.35 (.27-.42)	.45 (.38-.53)	.24 (.16-.32)
Surgeon B versus A, C-E	.45 (.39-.52)	.43 (.36-.51)	.36 (.28-.44)
Surgeon C versus AB, DE	.49 (.43-.56)	.31 (.23-.40)	.36 (.28-.44)
Surgeon D versus A-C, E	.42 (.35-.49)	.29 (.19-.39)	.29 (.20-.37)
Surgeon E versus A-D	.47 (.39-.54)	.38 (.30-.47)	.20 (.14-.27)

Table 1. Interobserver agreement between surgeons and peers *Continued*.

	Severity before treatment		**Results after treatment** [a]
	Severity [a]	*Symmetry* [a]	
Peers			
Overall agreement	.38 (.34-.41)		.22 (.19-.26)
Peer A versus B-E	.35 (.27-.43)		.23 (.15-.31)
Peer B versus A, C-E	.47 (.41-.53)		.22 (.14-.30)
Peer C versus AB, DE	.39 (.32-.46)		.23 (.16-.31)
Peer D versus A-C, E	.38 (.31-.46)		.30 (.23-.37)
Peer E versus A-D	.44 (.37-.51)		.23 (.15-.30)
Surgeons versus peers [b]	.54 (.48-.60)		.37 (.30-.45)

[a] K (95% CI)
[b] Arithmetic mean of surgeons versus arithmetic mean of peers

Table 2. Intraobserver agreement of peers and surgeons

	Severity before treatment		**Results after treatment** [a]
	Severity [a]	*Symmetry* [a]	
Surgeons			
Average agreement	.54 (.47-.61)	.60 (.46-.74)	.49 (.40-.58)
Surgeon A versus A	.40 (.28-.51)	.75 (.66-.84)	.50 (.37-.63)
Surgeon B versus B	.55 (.45-.64)	.63 (.52-.73)	.56 (.46-.67)
Surgeon C versus C	.59 (.41-.77)	.71 (.52-.89)	.61 (.41-.81)
Surgeon D versus D	.59 (.49-.68)	.34 (.21-.47)	.34 (.24-.45)
Surgeon E versus E	.55 (.45-.65)	.58 (.47-.69)	.44 (.34-.54)
Peers			
Average agreement	.48 (.38-.59)		.34 (.28-.40)
Peer A versus A	.34 (.23-.46)		.22 (.11-.34)
Peer B versus B	.61 (.53-.70)		.36 (.24-.47)
Peer C versus C	.50 (.40-.59)		.40 (.29-.50)
Peer D versus D	.38 (.28-.48)		.34 (.23-.44)
Peer E versus E	.60 (.51-.70)		.37 (.26-.37)

[a] K (95% CI)

Table 3. Average rating of severity before treatment and results after treatment

	Severity before treatment				Results after treatment			
	Overall	*DCS-bracing*	*Ravitch*	*P-value*	*Overall*	*DCS-bracing*	*Ravitch*	*P-value*
Surgeon A	1.67	1.60	1.84	.016	2.71	2.65	2.94	.002
Surgeon B	1.83	1.63	2.26	<.001	2.50	2.37	2.79	<.001
Surgeon C	1.97	1.76	2.44	<.001	2.33	2.12	2.79	<.001
Surgeon D	2.00	1.76	2.56	<.001	2.51	2.41	2.74	.004
Surgeon E	2.08	1.91	2.45	<.001	1.92	1.78	2.21	<.001
Peer A	2.33	2.22	2.60	<.001	2.34	2.30	2.44	.004
Peer B	2.21	2.01	2.66	<.001	2.33	2.38	2.21	.006
Peer C	1.84	1.65	2.26	<.001	2.05	2.01	2.15	.002
Peer D	1.96	1.83	2.24	.002	2.33	2.27	2.48	.020
Peer E	2.20	2.03	2.58	<.001	2.09	2.03	2.23	<.001

DCS-bracing = Dynamic Compression System Bracing

Table 4. Scars versus no scars

Surgeons	
Average agreement	.89 (.85-.94) [b]
Surgeon A	.90 (.81-.99) [a]
Surgeon B	.97 (.90-1.0) [a]
Surgeon C	.90 (.78-1.0) [a]
Surgeon D	.84 (.70-.98) [a]
Surgeon E	.86 (.75-.97) [a]
Average agreement	.39 (.19-.60) [b]
Peer A	.67 (.50-.83) [b]
Peer B	.27 (.14-.40) [b]
Peer C	.62 (.43-.80) [b]
Peer D	.20 (.06-.34) [b]
Peer E	.20 (.09-.31) [b]

[a] PABAK (95% CI)
[b] K (95% CI)

Difference in average ratings between the Ravitch and DCS-bracing group
Table 3 presents the differences in average ratings between the two treatment groups. Both surgeons and peers consistently rated deformities in patients treated with Ravitch surgery as more severe compared to those treated with DCS-bracing. The analysis of the results after treatment reveals a similar trend, with superior results in the Ravitch group compared to the DCS-bracing group. On average, peers rated deformities before treatment as more severe than surgeons (2.11 versus 1.91, P<.001). Peers on the other hand were less positive on the results compared to surgeons (2.23 versus 2.40, P<.001).

Influence of scar's, age and treatment duration on esthetic results
All surgeons exhibited an adequate and almost perfect agreement (κ=.89(.85-.94)) on results with scars compared to results 'imagining no scar', as shown in Table 4. This in contrast with patients' peers (κ=.39(.19-.60)), exhibiting inadequate agreement.

There was a significant relationship between age and severity of PC before treatment (surgeons and peers, P=.010, P=.029 resp.), but not between age and improvement after treatment (surgeons and peers, P=.205, P=.527 resp.).

There was no statistically significant relationship between ratings after treatment and duration of treatment (surgeons and peers, P=.682, P=.062 resp.) in de DCS-bracing group.

DISCUSSION

No adequate agreement was reached on esthetic results after treatment, severity of PC and symmetry. Surgeons exhibited better, but still inadequate intraobserver agreement compared to peers. Deformities of patients who underwent Ravitch surgery were regarded as more severe, but with better esthetic results afterwards compared to patients who underwent DCS-bracing. Peers rated chest deformities more negative than surgeons, assigning higher severity ratings before treatment and indicating less improvement afterward. Age was related to severity of PC before treatment. Both age and treatment duration were not related to final outcomes. More severe deformities before treatment obviously resulted in more noticeable improvements, as opposed to milder deformities.

Inter- and intraobserver agreement on results after treatment
While several studies report that treatment outcomes for pectus carinatum are determined based on patient satisfaction and mutually agreed-upon observed correction of the deformity,[13-15] our findings reveal that there was inadequate agreement

on esthetic outcomes post-treatment, with low intraobserver agreement indicating limited consistency among raters. Inconsistent assessment of esthetic outcomes can lead to unclear treatment endpoints, misaligned patient expectations and challenges in standardizing care. This underscores the need for objective evaluation methods in clinical practice.

A further complication is that developed pectoral muscles and a higher BMI can effectively mask PC. Many patients in our study with 'good improvement' developed big pectoral muscles, questioning whether PC actually improved, or was merely concealed by the pectoral muscles. If the results are primarily attributed to changes in muscle mass or BMI, there is a risk of recurrence of esthetic complaints when these factors change again.

Currently, patients are encouraged to build muscle as part of their treatment but are also cautioned that PC could reappear if they lose weight or muscle mass. Therefore, managing expectations before starting treatment is essential. Similar to observations in patients with pectus excavatum (PE) treated with vacuum bell therapy,[16] we find that as patients grow older and reach adulthood, they often become less concerned with their PC and are more likely to accept it if mild recurrence occurs over time.

Thus, an effective approach for obtaining a more objective assessment of PC outcomes should be one that is unbiased, but also capable of adjusting for variables in muscle mass and BMI to ensure accurate assessment of treatment success. 3D scanning is one such a technique,[6, 7, 17, 18, 19, 20, 21] and has been shown to be able to account for BMI.[22] This method can evaluate the initial severity of PC and, importantly, monitor progress throughout treatment. Over time, it may also serve to establish objective treatment endpoints, helping to create a more standardized approach to patient care.

However, in practice, some patients may feel satisfied earlier than others, regardless of set endpoints. Some patients may wish to end therapy before reaching the set endpoints, while others may prefer to continue beyond them. Despite having determined treatment endpoints, the cosmetic nature of PC means there are no risks associated with shorter or longer treatment durations, making these varying preferences a complication in the treatment process. Additionally, while 3D scanning is user-friendly, it demands extra time from physicians, whose time is already limited.

Inter- and intraobserver agreement on severity and symmetry before treatment
Determining which treatment is suitable for patients is usually decided based on pressure of initial correction (PIC), symmetry and flaring. When PC is too rigid or when the deformity is too asymmetrical or accompanied by flaring, a surgical correction is technically necessary.[5] Therefore the fair to moderate inter- and intraobserver agreement on PC severity is therefore less of a problem compared to the inconsistency regarding outcomes, since subjective evaluation of severity of PC does ultimately not influence the choice of treatment. Symmetry, on the contrary, does influence the choice of treatment, suggesting that the inadequate agreement on symmetry could have implications.

However, symmetry was not defined prior to the study because in practice also no consistent definition of symmetry was used. This may have led to the different interpretations of symmetry between surgeons. A clear, consistent definition of asymmetry is necessary since it can potentially influence clinical decision making. An asymmetry analysis as described in the literature could be utilized for this purpose.[17]

We found no relationship between severity of PC and treatment duration, likely due to inadequate inter- and intraobserver agreement on severity.

Ravitch versus DCS-bracing
Deformities in surgically treated patients were consistently rated as more severe than those in patients treated with DCS-bracing, by both surgeons and peers. Naturally, more severe deformities yield more noticeable improvements, compared to the milder deformities in the DCS-bracing group. This likely explains the better perceived results in the Ravitch group relative to those treated with DCS-bracing.

Interestingly, when examining the same data, peers tend to rate both the initial deformities and post-treatment results more negatively than surgeons. This may be due to surgeons' greater familiarity with these deformities, leading to lower severity ratings based on their clinical experience. Additionally, peers may be more sensitive to esthetic factors like scarring, which influences their assessments of post-treatment results more critically than those of surgeons.

Influence of scars on esthetic results after treatment
Our results show a significant difference in how surgeons and patients' peers perceive the impact of scars on results after treatment. Surgeons may regard scars as a minor issue due to their focus on achieving structural correction and clinical outcomes, viewing scars as a common, unavoidable aspect of effective surgical intervention.

This highlights the importance of considering individual perspectives when counseling patients on treatment options like the Ravitch procedure, and especially the minimally invasive Abramson procedure, as scars can significantly impact patients and are avoided with DCS-bracing.

Age and severity of treatment and influence on esthetic results

The relationship between age and severity of deformities before treatment may reflect the age difference between the Ravitch and DCS-bracing groups, with the Ravitch group generally being older. Patients in the Ravitch group also had more severe deformities, suggesting that severity might confound the observed association between age and deformity. While earlier bracing for younger patients could be considered to prevent deformity progression, we view the risk of relapse as a greater concern than progression and therefore prefer to initiate treatment at an older age to minimize this risk.[5] Additionally, age was not found to be related to better esthetic outcomes, suggesting there is no advantage to starting treatment earlier in terms of achieving improved results.

Influence of treatment duration on esthetic results

No relationship was found between treatment duration and results after treatment. This result might be biased because all our patients were already regarded as successfully treated.

Limitations

The fact that only 39.8% of patients had photographs available for analysis raises a concern regarding potential selection bias. While the reduced sample size may influence the results, there's no clear evidence that patients without photographs differ significantly in outcomes from those included in the analysis, since all patients were treated successfully and were satisfied with the treatment.

A limitation of our study is the potential bias introduced by the difference in experience between the peer and surgeon groups in assessing PC. However, this contrast was intentional, as it mirrors real-world scenarios where patients are inexperienced in evaluation of PC in contrast to surgeons. Understanding how these groups differ in their assessments provides valuable insights into patient experiences and expectations post-treatment. Additionally, the small number of peers reflects the exploratory nature of this experimental study, aimed at gaining preliminary insights rather than achieving broad generalizability.

Despite the impact of flaring on clinical decision making, a question regarding flaring was not included, because surgeons indicated that assessing flaring was not always feasible in our photographs. This would make analysis of flaring in our study unreliable. Although flaring is sometimes considered when choosing between surgery and bracing, its presence or absence does not impact treatment outcomes, which is the primarily focus of this study.

In our study we asked observers to visualize the photograph without scar, which may have introduced some bias by placing the burden of imagination on observers and potentially impacting the accuracy of their assessments. A more objective method, such as digitally removing scars from the photographs, could have helped to standardize evaluations and minimize bias by allowing observers to focus solely on the correction of the deformity itself. There may be a difference in visualization skills between peers and surgeons. However, given the clear distinction observed in their assessments, we believe our method proved effective overall.

Conclusion

Subjective assessments of pectus carinatum (PC) severity and treatment outcomes showed fair to moderate agreement, with peers rating deformities more severely and being more critical of scars. Ravitch surgery led to greater improvements for severe cases, but age and treatment duration had no significant impact on results. The need for objective tools like 3D scanning is clear to improve consistency and treatment evaluation. Future efforts should focus on refining these tools and addressing the psychosocial effects of scars in PC treatment.

REFERENCES

1 Welch KJ, Vos A. Surgical correction of pectus carinatum (pigeon breast). J Pediatr Surg. 1973;8:659-67.

2 Martinez-Ferro M, Bellia Munzon G, Fraire C, et al. Non-surgical treatment of pectus carinatum with the FMF(®) Dynamic Compressor System. J Vis Surg. 2016;2:57.

3 Martinez-Ferro M, Fraire C, Bernard S. Dynamic compression system for the correction of pectus carinatum. Semin Pediatr Surg. 2008;17:194-200.

4 Abramson H, Aragone X, Blanco JB, Ciano A, Abramson L. Minimally invasive repair of pectus carinatum and how to deal with complications. J Vis Surg. 2016;2:64.

5 Van Braak H, de Beer SA, Zwaveling S, Oomen MWN, De Jong JR. Ravitch Surgery or Dynamic Compression Bracing for Pectus Carinatum: A Retrospective Cohort Study. Ann Thorac Surg. 2024;117:144-152 .

6 Song SH, Kim CH, Moon DH, Lee S. Usefulness of 3-Dimensional Body Surface Scanning in the Evaluation of Patients with Pectus Carinatum. Korean J Thorac Cardiovasc Surg. 2020;53:301-5.

7 Belgacem A, Tricard J, Dutoit A, et al. Efficiency of non-operative management for pectus deformities in children using an X-ray-free protocol. Interdiscip Cardiovasc Thorac Surg. 2023;36.

8 De Beer SA, Blom YE, Lopez M, de Jong JR. Measured dynamic compression for pectus carinatum: A systematic review. Semin Pediatr Surg. 2018;27(3):175-82.

9 Emil S, Sévigny M, Montpetit K, et al. Success and duration of dynamic bracing for pectus carinatum: A four-year prospective study. J Pediatr Surg. 2017;52:124-9.

10 Light RJ. Measures of response agreement for qualitative data: Some generalizations and alternatives. Psychol Bull. 1971;76:365-77.

11 Byrt T, Bishop J, Carlin J. Bias, Prevalence and Kappa. J Clin Epidemiol. 1993;33:159-74 .

12 Landis JR, Koch GG. The Measurement of Observer Agreement for Categorical Data. Biometrics. 1977;33:159-74.

13 Dekonenko C, Dorman RM, Pierce A, et al. Outcomes Following Dynamic Compression Bracing for Pectus Carinatum. J Laparoendosc Adv Surg Tech A. 2019;29:1223-7.

14 Trinidad S, McKenna E, Jenkins T, et al. Outcomes From a Nurse Practitioner Led Dynamic Compression System Bracing Program for Pectus Carinatum. J Pediatr Surg. 2024;59:950-5.

15 Kelly RE, Obermeyer RJ, Goretsky MJ, et al. Ten-year experience with staged management of pectus carinatum: Results and lessons learned. J Pediatr Surg. 2021;56:1835-40.

16 Van Braak H, de Beer SA, al Ghouch Y, et al. 15 years of vacuum bell therapy for pectus excavatum: long-term outcomes and influencing factors. J Pediatr Surg. 2024.

17 Lain A, Garcia L, Gine C, Tiffet O, Lopez M. New Methods for Imaging Evaluation of Chest Wall Deformities. Front Pediatr. 2017;5:257.

18 Daemen JHT, Loonen TGJ, Lozekoot PWJ, et al. Optical imaging versus CT and plain radiography to quantify pectus severity: a systematic review and meta-analysis. J Thorac Dis. 2020;12:1475-87.

19 Wong KE, Gorton GE 3rd, Tashjian DB, Tirabassi MV, Moriarty KP. Evaluation of the treatment of pectus carinatum withh compressive orthotic bracing using three dimensional body scans. J Pediatr Surg. 2014;49:924-7.

20 Daemen JHT, de Loos ER, Geraedts TCM, et al. Visual diagnosis of pectus excavatum: An inter-observer and intra-observer agreement analysis. J Pediatr Surg. 2022;57:526-31.

21 Port E, Hebal F, Hunter CJ, Malas B, Reynolds M. Measuring the impact of brace intervention on pediatric pectus carinatum using white light scanning. J Pediatr Surg. 2018;53:2491-4 .

22 Taylor JS, Szafer D, Madhavan S, et al. Algorithm for Calculating Haller Index for Pectus Excavatum Using 3-Dimensional Optical Scanning. J Am Coll Surg. 2018;227:S192.

23 De Beer S, Volcklandt S, de Jong J, et al. Dynamic Compression Therapy for Pectus Carinatum in Children and Adolescents: Factors for Success. J Pediatr Surg. 2023;58:1440-5.

Chapter 5

Differences in chest wall between Dutch adolescents with pectus carinatum and the general pediatric population: a three-dimensional scanning analysis

Hendrik van Braak, Eelke Houter, Sjoerd A. de Beer, Sander Zwaveling, Matthijs W.N. Oomen, L.W. Ernest van Heurn, Justin R. de Jong

Journal of Pediatric Surgery. 2025. Online ahead of print.

ABSTRACT

Introduction

This study compares the External Haller Index in pectus carinatum patients to healthy adolescents, using 3D body imaging.

Methods

We conducted a cross-sectional study including pectus carinatum patients (aged 12–18) from Amsterdam UMC, and healthy adolescents from Sports Centre: Hoppers and the Jacobus Fruytier Scholengemeenschap, between December 2024 and May 2025. The External Haller Index was calculated using 3D scans (VECTRA®H2). Primary outcome was the difference in External Haller Index between pectus carinatum patients and healthy adolescents. Secondary outcomes included correlations between External Haller Index and influencing variables (gender, age, height, weight, Body Mass Index, Pressure of Initial Correction, complaints, syndromes, symmetry, flaring, scoliosis, previous treatment).

Results

Forty-three pectus carinatum patients and 50 healthy adolescents were included. Average age was fifteen years (14.0–16.0). Median External Haller Index was lower in pectus carinatum patients (1.4, IQR 1.3–1.4) than in healthy controls (1.6, IQR 1.5–1.7, P<.001), with a considerable overlap between groups. External Haller Index negatively correlated with Pressure of Initial Correction (P=−.36, P=.03) and was lower in patients with syndromes (P=.03). A positive correlation was found between External Haller Index and Body Mass Index (P=.22, P=.04). External Haller Index showed no association with reported complaints.

Conclusions

Pectus carinatum patients had significantly lower EHI than healthy peers, though overlap limits the use of a strict cut-off. This supports the use of 3D imaging as a non-invasive tool in PC assessment and highlight the need for Body Mass Index-adjusted reference values.

INTRODUCTION

Pectus carinatum (PC) is a congenital deformity of the anterior chest wall, characterized by the outward protrusion of the sternum, affecting approximately 0.3-0.7% of the population.[1, 5] It typically becomes apparent during the adolescent growth spurt, with patients primarily reporting chest pain, a distorted body image and low self-esteem.[3, 6, 7] Diagnosis is primarily based on clinical findings, although sometimes computed tomography (CT) is used to confirm the diagnosis.[8]

Orthotic bracing, particularly using the Dynamic Compression System (DCS), is the preferred treatment for PC.[9] However, treatment adherence can be challenging, and outcome assessment lacks standardization, often relying on subjective measures such as patient satisfaction.[10-12]

The Haller Index (HI), derived from imaging, is widely used to quantify sternal deformities and monitor treatment response.[7, 8, 11] Yet, repeated imaging raises concerns about radiation exposure in adolescents.[13, 14] As a safer alternative, three-dimensional (3D) body scanning allows for radiation-free assessment and enables calculation of the External Haller Index (EHI), which has shown promising results in evaluating pectus deformities and orthotic treatment outcomes.[7, 8, 15-18] However, EHI parameters for PC remain undefined, and normative reference values have not yet been established, as the majority of existing research has focused on pectus excavatum (PE).

This study aims to characterize the EHI values in patients with PC and to establish a normative range indicative of treatment success by comparing EHI values between healthy adolescents and those diagnosed with PC.

MATERIALS AND METHODS

Study design

A single-center cross-sectional study was conducted at the Department of Pediatric Surgery, Amsterdam UMC, location AMC, at Sports Centre: Hoppers, Hoorn and at a high school in Apeldoorn, the Netherlands (Jacobus Fruytier Scholengemeenschap).

Participants

Patients with PC (aged 12-18), regardless of severity, were enrolled during their scheduled visit to the outpatient clinic. Eligibility required that they had not undergone any prior intervention, including surgical procedures or orthotic bracing. Enrollment took place from December 2024, until May 2025. In addition to patients diagnosed with

PC, healthy adolescents (aged 12-18) were included in the study. These participants were assessed for chest wall deformities, and in cases of uncertainty, a second investigator was consulted for confirmation. Healthy adolescents were excluded if a chest wall deformity was present. Written informed consent was obtained from all participants and their parents or legal guardians prior to enrollment.

Treatment protocol and 3D imaging

Patients were treated using the FMF Dynamic Compression System brace (Pampamed®) according to our protocol (Figure 1). Prior to treatment, 3D body images were captured using the 3D scanning device VECTRA®H2, © 2024 Canfield Scientific, Inc..

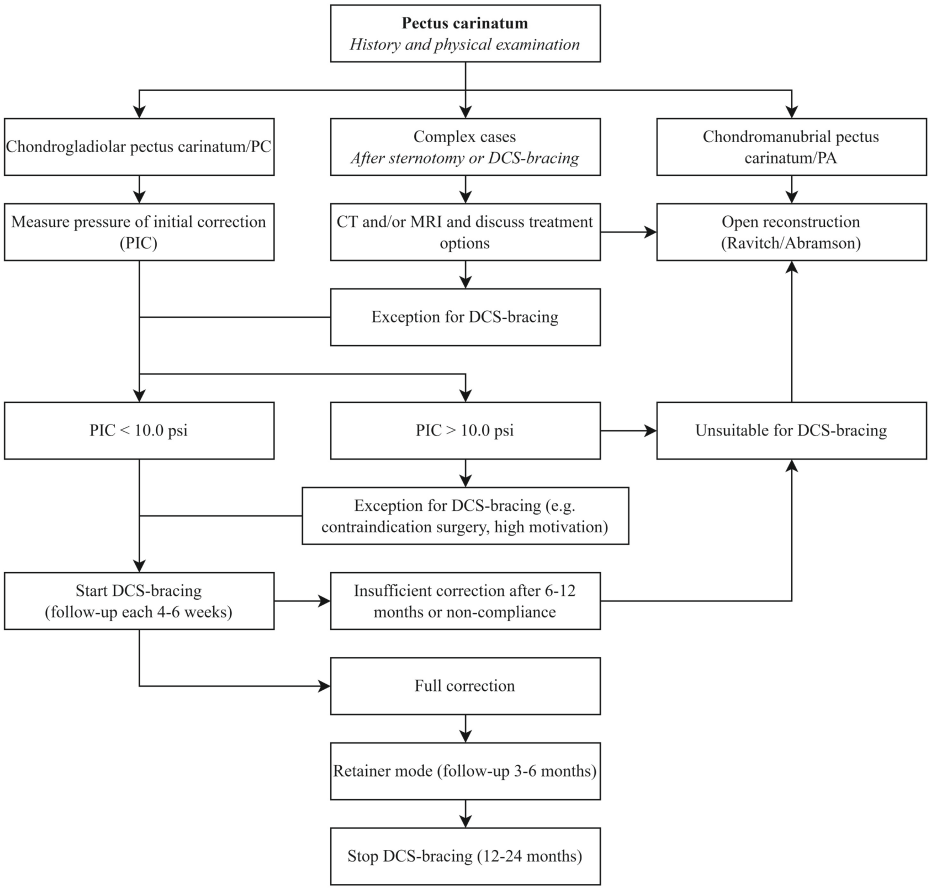


Figure 1 Treatment protocol DCS-bracing (CT = Computer Tomography, DCS = Dynamic Compression System, MRI = Magnetic Resonance Imaging, PA = Pectus Arcuatum, PC Pectus = Carinatum, PIC = Pressure of Initial Correction)

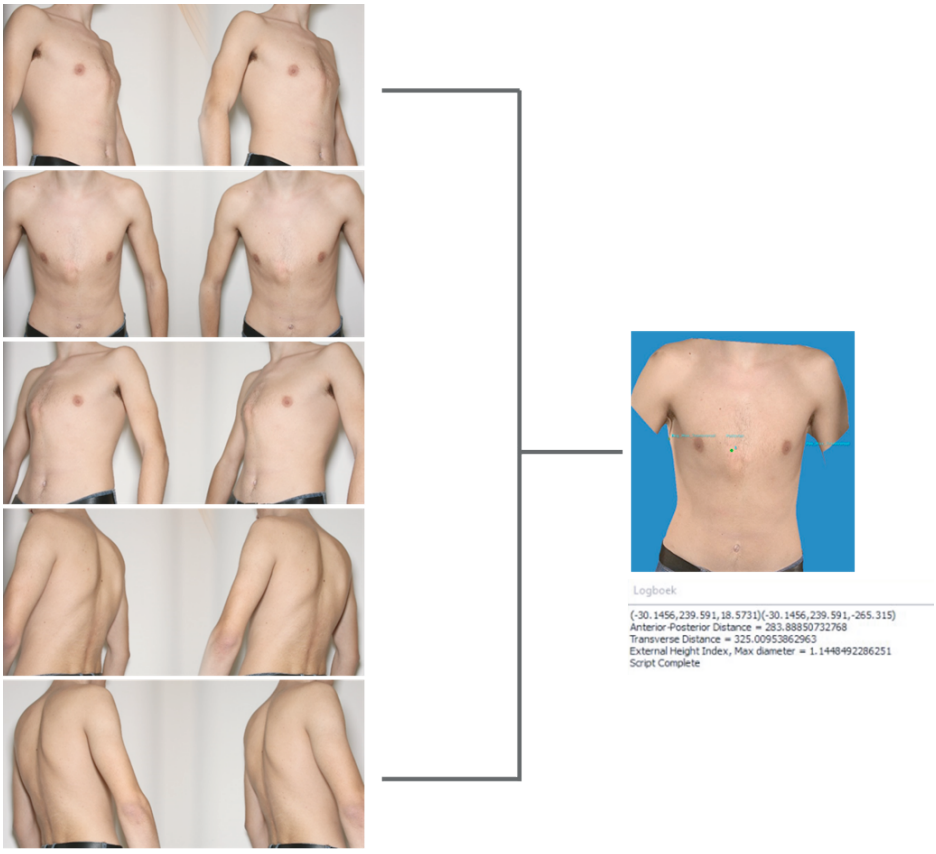


Figure 2 3D body images taken with Vectra H2 of a pectus carinatum patient

Standardized distance between patient and photographer was established by aligning two projected dots of the camera to coincide precisely on patient's chest. Patients are instructed to remove clothing covering the torso and stand upright with arms slightly extended and shoulders angled downward at 45°.

Scanning is performed at the end of exhalation, with patients briefly holding their breath.[4, 7, 19] Five images are captured to create a 360° view, with the photographer positioning the camera as follows: right and left lateral chest, angled upward from photographers hip level pointing toward just below the patient's chest; ventral chest, angled downward from the level of the photographer's nose toward the patient's chest; left and right dorsal sides, angled upward from photographers waist to a few centimeters above the patients lower edge of the shoulder blade. These angles ensure full torso exposure for accurate 3D imaging.[20] The full process takes less than five minutes per patient.

Measurements, outcomes and variables

Primary outcomes included the EHI in patients with PC and healthy adolescents. Secondary outcomes included the correlation between influencing factors and EHI (gender, age, height, weight, body mass index (BMI), PIC, complaints (shame, stress, reduced self-esteem, dyspnea or diminished physical endurance and pain), underlying conditions, (a)symmetry, flaring, scoliosis and previous treatment).

Data acquisition

Baseline characteristics were collected during the visit at the clinic. The PIC was measured by the surgeon during consultation. Patients were asked prior to scanning whether they had experienced psychosocial or somatic complaints. The response was documented as either 'yes' or 'no'. Patients were excluded whenever photos were not able to convert to a 3D scan, for instance due to inaccurate position of the patient or camera. The chest deformity was considered asymmetrical if the protrusion was not on the midline.

Calculation of EHI

The EHI was calculated by the Body Sculptor 3D aesthetic simulation software for VECTRA®H2, version 6.10, developed by Canfield Scientific, Inc.. The software was utilized to convert the raw image data of each patient into a 3D scan, demonstrated in Figure 2.[20] The software determines the EHI by first drawing a line from the highest point of deformity on the anterior side (positive direction) to the corresponding point on the posterior side, thereby determining the anterior-posterior (AP) distance of the body surface. Next, the software identifies the widest point of the body along this line and draws a perpendicular line at that site to measure the maximum body width. Finally, the EHI is automatically calculated by dividing the transverse diameter by the AP-distance of the body.[4, 9]

Statistical analysis

Statistical analyses were performed using IBM SPSS Statistics for Windows, Version 28.0 (Armonk, NY: IBM Corp.). Descriptive measurements were used to characterize the study population. Descriptive data were denoted as mean ± standard deviation (SD) for normally distributed data and the median and interquartile range (IQR) for skewed data. The Spearman's rank (Spearman's rho (P)) was used for correlation analyses. P-values were considered statistically significant at a level of <0.05.

RESULTS

Study population

Between December 2024 and May 2025, 43 PC patients and 50 healthy adolescents were included (Figure 3). Baseline characteristics are shown in Table 1 and Table 2. Of all patients, 90.3% (84/93) were male adolescents. The median age and BMI were respectively 15.0 (14.0–16.0) and 19.0 (17.9–20.3), mean height and weight were respectively 180.0 ± 9.2 cm and 62.0 ± 10.3 kg. Differences were found for age, weight and BMI between patients with pectus carinatum and healthy adolescents (respectively, P=.003, P=.001, P<.001).

Difference in EHI between PC and healthy adolescents

A significant difference in EHI between PC patients and healthy adolescents was found (P<.001). The median (IQR) EHI in PC patients was 1.4 (1.3–1.4) compared to 1.6 (1.5–1.7) in healthy adolescents. Figure 4 illustrates the measured EHIs with a noticeable degree of overlap in EHI between the groups.

Influence of different variables on the EHI.

Table 1 shows a negative relationship between PIC and EHI (P=-.36, P=.03), indicating that higher PIC values are associated with a lower EHI. A correlation analysis revealed a positive relationship between underlying diagnosis and EHI in PC patients (P=.03) with a median EHI of 1.2 for patients with a syndrome, including two patients with Marfan and one with Loeys-Dietz, in contrast to 1.4 for patients without syndromes. Furthermore, the analysis revealed a positive relationship between EHI and BMI across all participants (P=.22, P=.04), with higher BMI corresponding to an increase in EHI.

DISCUSSION

This study describes the EHI in PC patients and healthy adolescents using 3D body imaging. PC patients demonstrated a lower EHI compared to healthy adolescents. EHI values declined with increasing PIC and presence of a syndrome. Furthermore, a higher BMI correlated with an increased EHI.

EHI in PC patients

In current literature, the replacement of CT by 3D imaging has primarily been focused on PE patients.[15] The median EHI in our patients (1.4) aligns with previously reported EHI values of 1.3[21] and 1.5.[6, 18] Differences in EHI across studies may results from variations in scanning devices or body habitus.[22, 23]

Table 1 Baseline characteristics of pectus carinatum patients and the influence of these characteristics on EHI

	Pectus carinatum (n=43)	P-value
Sex [a]		.32
Male	40 (93)	
Female	3 (7)	
Age (years) [b]	15.0 (14.0–16.0)	.06
Height (cm)	178 ± 8.0	.80
Weight (kg)	59.6 ± 8.8	.68
BMI (kg/m^2) [b]	18.6 (17.6–19.8)	.27
EHI [b]	1.4 (1.3–1.4)	
PIC (psi)	6.6 ± 2.1	.03
Symmetry [a]	18 (42)	.36
Scoliosis [a]	7 (16)	.17
Flaring [a]	7 (16)	.57
Underlying diagnosis [a]	3 (9)	.03
Somatic complaints [a]	10 (23)	.82
Psychological complaints [a]	2 (5)	.15
Previous treatment [a]	3 (7)	.74

BMI = Body Mass Index, EHI = External Haller Index, IQR = Interquartile Range, n = number, PIC = Pressure Initial Compression, psi = pounds per square inch, SD = Standard Deviation.
[a] Data displayed as n (%)
[b] Continues variables expressed as median (IQR); all remaining continuous variables are expressed as mean ± standard deviation

Table 2 Baseline characteristics of healthy patients and the influence of these characteristics on EHI

	Healthy adolescents (n=50)	P-value
Sex [a]		.87
Male	44 (88)	
Female	6 (12)	
Age (years) [b]	16.0 (15.0–16.0)	.88
Height (cm)	179 ± 10	.21
Weight (kg)	66.0 ± 10.7	.85
BMI (kg/m^2) [b]	20.1 (18.9–21.7)	.78
EHI [b]	1.6 (1.5–1.7)	

BMI = Body Mass Index, EHI = External Haller Index, IQR = Interquartile Range, n = number, SD = Standard Deviation.
[a] Data displayed as n (%)
[b] Continues variables expressed as median (IQR); all remaining continuous variables are expressed as mean ± standard deviation

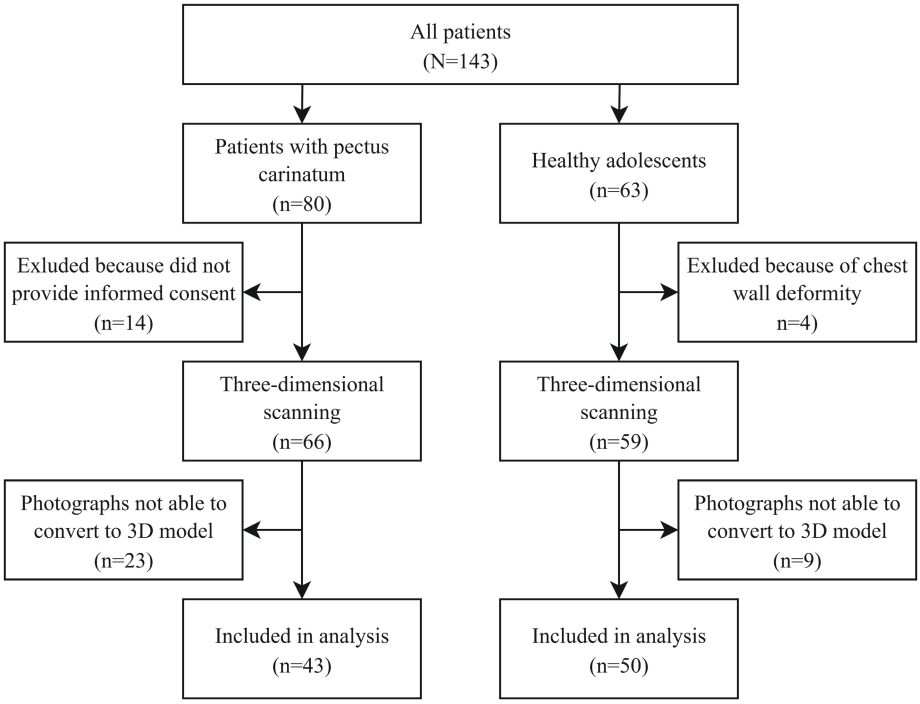


Figure 3 Inclusion flowchart of participants

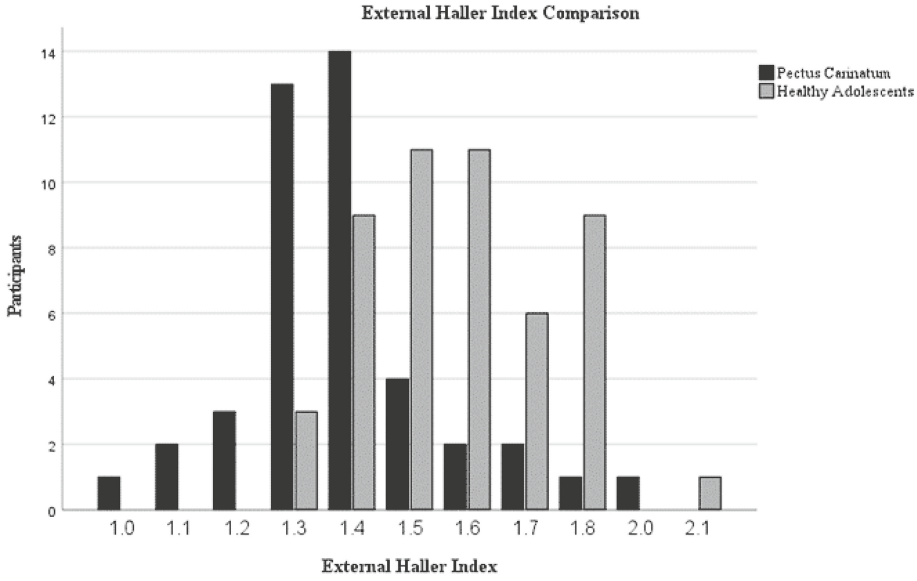


Figure 4 External Haller Index in pectus carinatum patients and healthy adolescents

The difference in EHI between PC patients and healthy controls.
A difference in EHI was observed between PC patients and healthy adolescents, with a considerably lower EHI in PC patients. This suggests standardized treatment outcomes for PC patients can be established, potentially leading to a uniform assessment of DCS-bracing. Given the overlap (Figure 4), defining values as a range rather than a single threshold may be more appropriate. However, due to the limited sample size, our current data are insufficient to establish such a range, and further studies with larger cohorts are needed to determine reliable cut-off values.

The overlap in EHI between healthy patients and PC patients suggests that the EHI may not be the only outcome parameter for treatment success. Some patients with an EHI within the normal range still seek treatment, indicating that their condition is perceived as abnormal despite falling within standard parameters. This highlights the significant role of aesthetic concerns in decision-making for initiating treatment.[5] On the other hand, overtreatment may occur, as some individuals may undergo unnecessary treatment despite having a chest within normal range. These patients might benefit more from alternative approaches, such as strength training or posture-improving exercises.

Influence of various variables on EHI
This may be particularly relevant, as our study did not find a significant correlation between the EHI and patient complaints, indicating that symptom severity does not necessarily reflect the extent of the deformity. Our results did however demonstrate a relationship between EHI and PIC, where a higher PIC was associated with a lower EHI, thus more severe deformity.[10]

According to the literature, PC is in some cases associated with certain syndromes.[24, 25] While our results appear to support this association, the presence of only three patients with a syndrome in our cohort limits the strength of this finding. Therefore, this association requires validation in future studies involving larger cohorts.

In addition, we found an association between EHI and BMI, with a higher BMI corresponding with increasing EHI values. However, in this study no adjustment for body habitus was applied, as the formula proposed by Taylor et al. was deemed inapplicable in our PC population, and no formula for adjustment for BMI has yet been established for PC patients.[23]

Limitations

Establishing a control group remains challenging, primarily due to the limited commitment for participation among adolescents. In our study, our healthy controls had a higher median body weight compared to the PC patients, which potentially affect EHI values, as no corrections for weight were made. This is important to highlight, as it raises the question of whether BMI acts as a confounder for EHI, suggesting that future analyses should adjust for weight or BMI.

Acquiring images for 3D scanning protocol revealed a learning curve throughout the study, as 32 participants were excluded from the analysis due to non-convertibility of the photographs. The process of capturing 3D images depends on factors such as body constitution, positioning, and camera angle.[22]

Correlation analysis between 3D scanning and CT scanning was not conducted, as CT scans were not performed, given this is not standard protocol and due to its previous demonstrated comparability in correlation studies.[8, 15] Besides, previous literature has demonstrated its utility in monitoring DCS-bracing.[17, 18, 26, 27] Instead, research should primarily focus on establishing reference parameters to enhance follow-up and to improve the key determinant of DCS-bracing success: motivation.[16, 24]

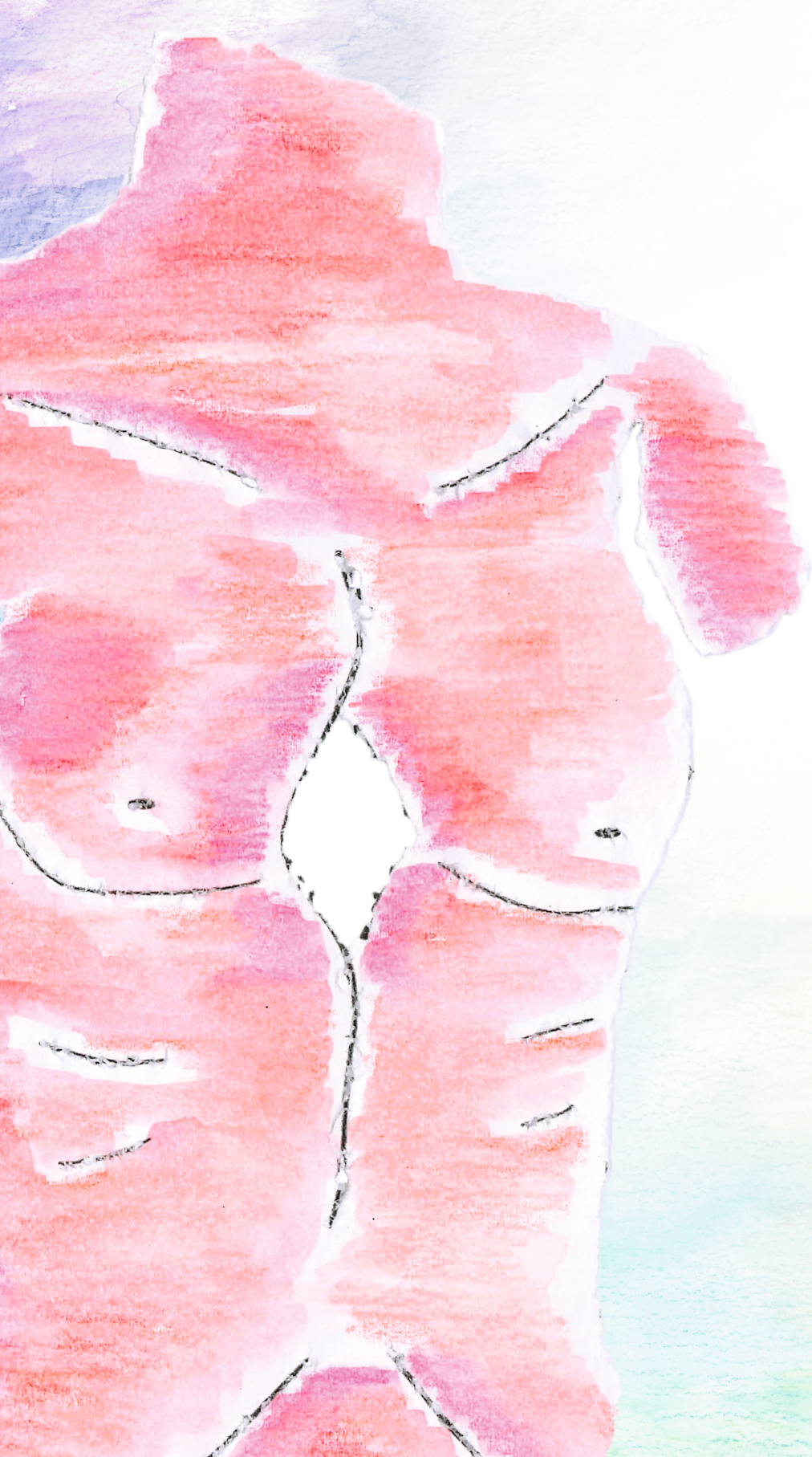
Conclusion

This study, to our knowledge, is the first in current literature to compare EHI values between PC patients and healthy adolescents at this extent. This study demonstrated a significantly lower EHI compared to healthy adolescents. EHI values in PC patients decreased with increasing PIC and with the presence of a syndrome. Future studies should consider correction of the EHI for weight or BMI. Establishing reference parameters is crucial for standardizing treatment outcomes of DCS-bracing in the future. Our findings provide promising results, contributing the potential use of 3D body imaging as a radiation-free alternative to develop standardized treatment outcomes.

REFERENCES

1. Robicsek F, Watts LT. Pectus carinatum. Thorac Surg Clin. 2010;20(4):563-74.
2. Park CH, Kim TH, Haam SJ, Lee S. Does overgrowth of costal cartilage cause pectus carinatum? A three-dimensional computed tomography evaluation of rib length and costal cartilage length in patients with asymmetric pectus carinatum. Interact Cardiovasc Thorac Surg. 2013;17(5):757-63.
3. Janssen N, Coorens NA, Franssen A, et al. Pectus excavatum and carinatum: a narrative review of epidemiology, etiopathogenesis, clinical features, and classification. J Thorac Dis. 2024;16(2):1687-701.
4. Servi M, Buonamici F, Furferi R, et al. Pectus Carinatum: a non-invasive and objective measurement of severity. Med Biol Eng Comput. 2019;57(8):1727-35.
5. De Beer SA, Blom YE, Lopez M, de Jong JR. Measured dynamic compression for pectus carinatum: A systematic review. Semin Pediatr Surg. 2018;27(3):175-82.
6. Orrick BA, Pierce AL, McElroy SF. Changes in self-image after pectus carinatum brace treatment. J Pediatr Surg. 2022;57(8):1579-83.
7. Guillot MS, Rouchaud A, Mounayer C, et al. X-ray-free protocol for pectus deformities based on magnetic resonance imaging and a low-cost portable three-dimensional scanning device: a preliminary study. Interact Cardiovasc Thorac Surg. 2021;33(1):110-8.
8. Belgacem A, Tricard J, Dutoit A, et al. Efficiency of non-operative management for pectus deformities in children using an X-ray-free protocol. Interdiscip Cardiovasc Thorac Surg. 2023;36(6).
9. Van Braak H, de Beer SA, Zwaveling S, Oomen MWN, de Jong JR. Ravitch Surgery or Dynamic Compression Bracing for Pectus Carinatum: A Retrospective Cohort Study. Ann Thorac Surg. 2024;117(1):144-50.
10. Martinez-Ferro M, Bellia Munzon G, Fraire C, et al. Non-surgical treatment of pectus carinatum with the FMF((R)) Dynamic Compressor System. J Vis Surg. 2016;2:57.
11. Desmarais TJ, Keller MS. Pectus carinatum. Curr Opin Pediatr. 2013;25(3):375-81.
12. Martinez-Ferro M, Fraire C, Bernard S. Dynamic compression system for the correction of pectus carinatum. Semin Pediatr Surg. 2008;17(3):194-200.
13. Miglioretti DL, Johnson E, Williams A, et al. The use of computed tomography in pediatrics and the associated radiation exposure and estimated cancer risk. JAMA Pediatr. 2013;167(8):700-7.
14. Don S. Radiosensitivity of children: potential for overexposure in CR and DR and magnitude of doses in ordinary radiographic examinations. Pediatr Radiol. 2004;34 Suppl 3:S167-72; discussion S234-41.
15. Daemen JHT, Loonen TGJ, Lozekoot PWJ, et al. Optical imaging versus CT and plain radiography to quantify pectus severity: a systematic review and meta-analysis. J Thorac Dis. 2020;12(4):1475-87.
16. Lain A, Garcia L, Gine C, Tiffet O, Lopez M. New Methods for Imaging Evaluation of Chest Wall Deformities. Front Pediatr. 2017;5:257.
17. Song SH, Kim CH, Moon DH, Lee S. Usefulness of 3-Dimensional Body Surface Scanning in the Evaluation of Patients with Pectus Carinatum. Korean J Thorac Cardiovasc Surg. 2020;53(5):301-5.
18. Wong KE, Gorton GE, 3rd, Tashjian DB, Tirabassi MV, Moriarty KP. Evaluation of the treatment of pectus carinatum with compressive orthotic bracing using three dimensional body scans. J Pediatr Surg. 2014;49(6):924-7.

19. Uccheddu F, Ghionzoli M, Volpe Y, et al. A Novel Objective Approach to the External Measurement of Pectus Excavatum Severity by Means of an Optical Device. Ann Thorac Surg. 2018;106(1):221-7.
20. Canfield Scientific I. VECTRA®H2 User Guide. Parsippany (NJ); 2020.
21. Poncet P, Kravarusic D, Richart T, et al. Clinical impact of optical imaging with 3-D reconstruction of torso topography in common anterior chest wall anomalies. J Pediatr Surg. 2007;42(5):898-903.
22. Daemen JHT, Loonen TGJ, Verhulst AC, et al. Three-Dimensional Imaging of the Chest Wall: A Comparison Between Three Different Imaging Systems. J Surg Res. 2021;259:332-41.
23. Taylor JS, Madhavan S, Szafer D, et al. Three-Dimensional Optical Imaging for Pectus Excavatum Assessment. Ann Thorac Surg. 2019;108(4):1065-71.
24. De Beer S, Volcklandt S, de Jong J, et al. Dynamic Compression Therapy for Pectus Carinatum in Children and Adolescents: Factors for Success. J Pediatr Surg. 2023;58(8):1440-5.
25. Notrica DM, McMahon LE, Jaroszewski DE. Pectus Disorders: Excavatum, Carinatum and Arcuatum. Adv Pediatr. 2024;71(1):181-94.
26. Szafer D, Taylor JS, Pei A, et al. A Simplified Method for Three-Dimensional Optical Imaging and Measurement of Patients with Chest Wall Deformities. J Laparoendosc Adv Surg Tech A. 2019;29(2):267-71.
27. Port E, Hebal F, Hunter CJ, Malas B, Reynolds M. Measuring the impact of brace intervention on pediatric pectus carinatum using white light scanning. J Pediatr Surg. 2018;53(12):2491-4.



Part II

Pectus excavatum

Chapter 6

15 years of vacuum bell therapy for pectus excavatum: long-term outcomes and influencing factors

Hendrik van Braak, Sjoerd A. de Beer, Youssef al Ghouch, Sander Zwaveling, Matthijs W.N. Oomen, L.W. Ernest van Heurn, Justin R. de Jong

Journal of Pediatric Surgery. 2025 Feb 60:161891.

ABSTRACT

Background

Long-term results and factors affecting outcomes of vacuum bell therapy for pectus excavatum are relatively unknown.

Methods

We conducted a retrospective study on patients (<18y) treated with vacuum bell therapy between May 2008 and October 2021. Primary outcome was treatment success; secondary outcomes were analysis of daily time spent on treatment, treatment duration, complications, long-term follow-up, treatment for patients awaiting a Nuss procedure, treatment for female patients, and factors affecting outcomes.

Results

Of 259 patients treated with vacuum bell therapy, 18.9% (49/259) were still being treated, 17.4% (45/259) were lost to follow-up and 63.7% (165/259) completed treatment, with a 52.1% (86/165) success rate. Median follow-up was 64.0 months (interquartile range 48.0-87.0). More time spent daily on vacuum bell therapy, total treatment duration, and overnight use led to a higher success rate (P=.002, P<.001, P<.001 resp.). Complications (22.8%, 59/259) were minor, recurrence occurred in 2.3% (2/86) of patients. Of the patients treated while awaiting a Nuss procedure, 26.7% (4/15) no longer required the Nuss procedure. Breast growth made 39.3% (11/28) of female patients quit treatment. Deeper deformities (P=.02, P=.009), flexible chest wall (P=.007) and symptomatic pectus excavatum (P=.02) resulted in lower success rates.

Conclusions

Vacuum bell therapy is successful in up to 52.1% of patients. Overnight vacuum bell use and treatment while awaiting a Nuss procedure should be encouraged. Older patients with a stiff chest wall can be successfully treated with prolonged treatment. For female patients watchful waiting or early treatment, to prevent challenges during breast growth, is preferred.

INTRODUCTION

Pectus excavatum (PE) is a chest wall anomaly with an incidence of 0.1-0.8%, predominantly affecting males (2:1 to 9:1 ratio). PE is usually discovered during or at the start of puberty.[1] Patients may suffer from physical complaints, but also experience psychological distress due to a disturbed body image.[2, 3]

Minimally invasive repair of PE is currently the most common treatment.[4] The results are usually excellent, however sometimes serious complications occur.[5, 6] An alternative non-surgical treatment option is vacuum bell (VB) therapy, which is usually used in patients with relatively mild deformities.[7] VB therapy is effective for treating PE; however, the treatment period is long, and a relatively large number of patients continue active treatment thereafter.[8] Additionally, in recent literature percentages of success vary, and high percentages of non-compliance (9.7%-36.1%) are reported.[9-12] Furthermore, most studies comprise small numbers of patients and lack long-term follow-up, which is needed to evaluate the long-term results and the risk of recurrence. Numerous studies have tried to identify factors that may contribute to success or compliance, but the outcomes vary.[11, 13-16]

We share our 15 years' experience with VB therapy in the Amsterdam Pectus Center, focusing on long-term outcomes and factors influencing the treatment outcomes.

PATIENTS AND METHODS

Study population and design

We performed a retrospective cohort study of patients (<18 years) undergoing VB (Eckart Klobe Vacuum Bell) therapy in the Amsterdam Pectus Center between May 2008 and October 2021. A Medical Ethics Review Committee (METC) official waiver of ethical approval was granted by the METC of the Amsterdam Medical Center. Informed consent was obtained from all patients (or their parents).

Data extraction

Data were retrospectively obtained from patient records. Patients who had not visited or contacted the outpatient clinic for over twelve months, without completing their treatment, were contacted by phone; those unreachable were considered lost to follow-up.

Overnight hours were excluded from the calculation of daily time spend on VB therapy, because some patients mention that the VB detaches during the night while they are asleep, and it is unclear at which time point this occurs, making it impossible to count these hours. Therefore nightly use was included as a separate, nominal variable. Chest wall flexibility was assessed by attaching the VB to the patient's chest, applying suction, and subsequently assessing sternal elevation. Flexibility was deemed present when sternal elevation occurred effectively within minutes. Sternal depression was measured using the method described by Obermeyer et al.[10]

Treatment protocol

After anamnesis and physical examination, patients are offered the choice between the Nuss procedure and VB therapy. The Nuss procedure is typically recommended, and we explain that VB therapy is most effective in cases of mild to moderate PE, as assessed by the physician based on sternal depression and visual evaluation. If the patient or their parents prefer to avoid surgery, or if the patient is not eligible for the Nuss procedure due to very mild PE, VB therapy is advised. Before starting VB therapy, patients undergo a trial fitting of the VB to evaluate their suitability for treatment.

X-rays or CT scans are not routinely performed unless pectus arcuatum is suspected by the clinician. Initially, our protocol included conducting electrocardiograms and echocardiograms to assess for cardiac abnormalities before starting VB therapy. However, after determining that cardiac abnormalities did not impact the effectiveness of VB therapy, we removed these tests from the standard procedure, except in severe symptomatic cases where patients are referred for cardiac and pulmonary evaluations.

Patients are instructed to begin using the VB two to three times per day for 30 to 60 minutes, gradually increasing the duration and the applied suction as tolerated. We recommend wearing the VB overnight and during routine activities such as eating, homework, or gaming, to help accumulate the necessary wearing hours. However, it is important to note that overnight use should not begin in the first two to three months, since the skin has to adapt in order to endure the prolonged suction applied overnight. There is no standard treatment duration; if there is still progression and the patient remains motivated, we advise them to continue using the VB. After the chest wall is fully corrected, patients enter the retainer phase, i.e., the treatment phase in which wearing time is gradually reduced to maintain chest wall correction. Treatment is considered successful if patients, parents and surgeon regard the result as aesthetically pleasing and VB therapy is stopped.

While we aim to offer VB therapy to all patients on the waiting list for the Nuss procedure, financial constraints prevent us from doing so.

Outcomes

The primary outcome of the study was percentage of patients which were successfully treated; secondary outcomes consisted of analysis of daily time spent on VB therapy, treatment duration, complications, long-term follow-up, VB therapy for patients awaiting the Nuss procedure, VB therapy for female patients, and prognostic factors influencing the treatment (familiar predisposition, sex, age, length, weight, Body Mass Index (BMI), comorbidities, flexibility chest, symmetry, flaring, scoliosis, symptomatic PE, increase during puberty, use of VB after treatment for pectus carinatum (PC), sternal depression, type of VB, frequency of wearing, wearing at night, complications).

Statistical analysis

Descriptive measurements were utilized to characterize the study population. Statistical significance was set at $P<0.05$. Data were analyzed using IBM SPSS Statistics 28.0.

RESULTS

All (N=259) patients who started VB therapy in the study period were included in the analysis. Median age was fifteen (13.0-16.0) years and 89.2% of patients were male (231/259). Median follow-up was 64.0 (interquartile range (IQR) 48.0-87.0) months. Baseline characteristics are described in Table 1. Eight patients suffered from cardiovascular comorbidities (supraventricular tachycardia (n=2), atrial septal defect (n=1), ventricular septal defect (n=1), cardiac souffle (n=2) and bradycardia (n=2)). Twenty-seven patients suffered from respiratory comorbidities (asthma (n=12), allergic rhinitis (n=8), hyperreactive airway disease (n=5), history of bilateral pneumothorax (n=1) or bronchiolitis (n=1)). Twelve patients were treated for chest wall deformities in the past (PC treated with Ravitch (n=5) or the Dynamic Compression System (DCS) (n=2), PE treated with the Nuss procedure (n=3)), or had chest wall deformities (Poland syndrome (n=1) and costal flaring (n=1)). Characteristics of PE deformities and symptoms are described in Table 2. Treatment characteristics and outcomes are described in Table 3 and Table 4.

Success of treatment and loss to follow-up

At the time of analysis, 18.9% (49/259) of patients were actively treated, 17.4% (45/259) were lost to follow-up and 63.7% (165/259) had finished treatment. Of the latter group, 52.1% (86/165) had finished treatment successfully (Figure 1).

Of the patients lost to follow-up, 35.6% (16/45) were lost before their six-month check-up and 68.9% (31/45) before their eighteen-month check-up. The median treatment duration until the final check-up was ten months (IQR 2.0-18.0).

Daily time spent on VB therapy, treatment duration

The average, minimum and maximum daily time of wearing the VB was higher in the successfully treated group (3.0 (IQR 2.0-4.3) versus 2.0 (IQR 1.3-3.0), 2.0 (IQR 1.0-3.0) versus 1.5 (IQR 1.0-2.0) and 3.5 (IQR 2.5-6.0) versus 3.0 (IQR 2.0-5.0), respectively). All were related to a higher success rate (P=.002, P=.004, P=.01, respectively). Total treatment duration was longer in successfully treated patients (24.0 (IQR 20.0-38.0) compared to 13.5 (IQR 8.0-26.0), P<.001)) in unsuccessfully treated patients.

Complications and long-term follow-up

Of all patients 22.8% had one or more complications (Table 3). All complications were minor, mainly skin-related and did not affect treatment success (P=.17). Pain (P=.03) and other complications (motivation problems (n=5), acne (n=2), thickened glandular disk (n=3) and shortness of breath (n=2)) (P=.04) were more prevalent in the unsuccessfully treated group. There were no significant long-term complications. Of all patients who were treated unsuccessfully, 45.6% (n=36/79) underwent a Nuss procedure.

In 2.3% (2/86) of patients who were initially treated successfully, PE recurred. One of them was treated at a young age (11y) and experienced recurrence when his growth spurt began.

VB therapy for patients awaiting the Nuss procedure

Not every patient awaiting the Nuss procedure received VB therapy due to financial constraints. Of the patients that did start with VB therapy, 53.3% (8/15) did not undergo a Nuss procedure yet. Two of them were lost to follow-up, two were unsuccessfully treated with VB therapy and refrained from a Nuss procedure, two patients finished treatment successfully and two patients are still being treated with VB therapy.

Table 1. Patient characteristics

	All patients *(N=259)*	**Patients who finished treatment** *(n=165)*		**P-value**
		Successful (n=86)	*Unsuccessful (n=79)*	
Sex [b]				.78
Male	231 (89.2)	75 (87.2)	70 (88.6)	
Female	28 (10.8)	11 (12.8)	9 (11.4)	
Age [a]	15.0 (13.0-16.0)	15.0 (14.0-16.0)	14.0 (13.0-16.0)	.22
Length (cm) (n=140) [a]	175.0 (167.0-182.8)	175.0 (167.5-182.5)	174.5 (165.8-180.0)	.33
Weight (kg) (n=136) [a]	57.0 (47.3-65.0)	58.0 (49.3-65.0)	53.8 (45.8-62.3)	.22
BMI (n=131) [a]	17.7 (16.1-19.3)	17.8 (15.7-19.7)	17.7 (15.9-18.7)	.51
Follow-up (months) [a]	64.0 (48.0-87.0)	74.0 (54.8-90.3)	64.0 (50.0-84.0)	.10
Family history PE/PC [b]	95 (36.7)	37 (43.0)	29 (36.7)	.48
Comorbidities [b]	110 (42.5)	39 (45.3)	35 (44.3)	.89
Type of comorbidities [b]				
Cardiovascular	8 (3.1)	4 (4.7)	1 (1.3)	.21
Respiratory	27 (10.4)	10 (11.6)	11 (13.9)	.66
Gastrointestinal	9 (3.5)	3 (3.5)	2 (2.5)	.72
Chest wall deformities	12 (4.6)	3 (3.5)	5 (6.3)	.40
Other musculoskeletal	18 (6.9)	6 (7.0)	5 (6.3)	.87
Cutaneous	16 (6.2)	7 (8.1)	6 (7.6)	.90

Table 1. Patient characteristics *Continued.*

	All patients *(N=259)*	**Patients who finished treatment** *(n=165)*		**P-value**
		Successful (n=86)	*Unsuccessful (n=79)*	
Other	42 (16.2)	14 (16.3)	15 (19.0)	.65
Abnormalities electrocardiogram (n=81) [b]	17 (6.6)	9 (10.5)	5 (6.3)	.34
Abnormalities echocardiogram (n=83) [b]	9 (3.5)	4 (4.7)	3 (3.8)	.79

[a] Continuous variables expressed as median (IQR)
[b] Data displayed as n (%)
BMI = Body Mass Index, PC = Pectus Carinatum, PE = Pectus excavatum

Table 2. Pectus excavatum characteristics, symptoms

	All patients *(N=259)*	**Patients who finished treatment** *(n=165)*		**P-value**
		Successful (n=86)	*Unsuccessful (n=79)*	
After treatment for PC [b]	10 (3.9)	5 (5.8)	2 (2.5)	.30
Increase of PE during puberty [b]	140 (54.1)	46 (53.5)	43 (54.4)	.90
Flaring [b]	110 (42.5)	39 (45.3)	28 (35.4)	.20
Scoliosis [b]	32 (12.4)	11 (12.8)	9 (11.4)	.78
Symmetrical [b]	203 (78.4)	22 (25.6)	11 (13.9)	.06
Flexible [b]	132 (51.0)	33 (38.4)	47 (59.5)	.007
Sternal depression (cm) (n=67) [a]	2.0 (1.5-2.5)	1.8 (1.5-2.0)	2.1 (1.9-2.5)	.02
Sternal depression (narrative) [b]				.009
Mild/light	63 (24.3)	38 (44.2)	21 (26.6)	
Moderate	104 (40.2)	34 (39.5)	30 (38.0)	
Deep/severe	92 (35.5)	14 (16.3)	28 (35.4)	
Symptomatic [b]	120 (46.3)	34 (39.5)	46 (58.2)	.02
Pain/stabbing pain	65 (25.1)	18 (20.9)	21 (26.6)	.39
Pressure on chest	20 (7.7)	7 (8.1)	5 (6.3)	.66
Palpitations	12 (4.6)	3 (3.5)	5 (6.3)	.40
Endurance problems	69 (26.6)	16 (18.6)	29 (36.7)	.009
Appearance/shame [b]	135 (52.1)	38 (44.2)	44 (55.7)	.14

[a] Continuous variables expressed as median (IQR)
[b] Data displayed as n (%)
PC = Pectus Carinatum, PE = Pectus excavatum

Table 3. Treatment characteristics

	All patients *(N=259)*	**Patients who finished treatment** *(n=165)*		**P-value**
		Successful (n=86)	*Unsuccessful (n=79)*	
Type of VB [b]				.08
Mini	62 (23.9)	23 (26.7)	20 (25.3)	
Small	149 (57.5)	43 (50.0)	48 (60.8)	
Bodybuilder	26 (10.0)	13 (15.1)	5 (6.3)	
Woman	15 (5.8)	7 (8.1)	3 (3.8)	
Big	7 (2.7)	0 (.0)	3 (3.8)	
Average daily time of VB therapy (h) (n=225) [a]	2.5 (1.6-4.0)	3.0 (2.0-4.3)	2.0 (1.3-3.0)	.002
Minimum daily time (h) (n=228)	2.0 (1.0-2.5)	2.0 (1.0-3.0)	1.5 (1.0-2.0)	.004
Maximum daily time (h) (n=227)	3.0 (2.0-6.0)	3.5 (2.5-6.0)	3.0 (2.0-5.0)	.01
Frequency of wearing (daily) (n=233) [a]	1.0 (1.0-2.0)	2.0 (1.0-2.0)	1.0 (1.0-2.0)	.16
Minimal frequency (n=231)	1.0 (1.0-2.0)	1.0 (1.0-2.0)	1.0 (1.0-2.0)	.13
Maximal frequency (n=231)	2.0 (1.0-3.0)	2.0 (1.0-3.0)	2.0 (1.0-3.0)	.08
At night/during sleep [b]	119 (45.9)	50 (58.1)	24 (30.4)	<.001
Complications during treatment [b]	59 (22.8)	21 (24.4)	27 (34.2)	.17

Table 3. Treatment characteristics *Continued.*

	All patients *(N=259)*	**Patients who finished treatment** *(n=165)*		**P-value**
		Successful (n=86)	*Unsuccessful (n=79)*	
Type of complications [b]				
Skin discoloration	29 (11.2)	13 (15.1)	9 (11.4)	.48
Pain	16 (6.2)	3 (3.5)	10 (12.7)	.03
Ulcers/blisters	5 (1.9)	2 (2.3)	1 (1.3)	.61
Wounds	4 (1.5)	1 (1.2)	1 (1.3)	.95
Edema	5 (1.9)	1 (1.2)	2 (2.5)	.51
Other	12 (4.6)	2 (2.3)	8 (10.1)	.04

[a] Continues variables expressed as median (IQR)
[b] Data displayed as n (%)
VB = vacuum bell, h = hours

Table 4. Outcomes of patients who finished treatment

	All patients who finished treatment	**Successful**	**Unsuccessful**	**P-value**
	(n=165)	*(n=86)*	*(n=79)*	
Start retainer phase after (months) [a]	21.0 ± 10.1	21.8 ± 10.7	.	.
Average retainer phase duration (months)	10.0 (6.0-17.0)	10.0 (6.0-17.0)	.	.
Total wearing time (months)	23.0 (12.0-32.5)	24.0 (20.0-38.0)	13.5 (8.0-26.0)	<.001
Age at end of treatment	16.0 (15.0-18.0)	17.0 (16.0-18.0)	16.0 (14.0-17.0)	<.001
Nuss after VB [b]	36 (21.8)	0 (.0)	36 (45.6)	<.001
Retreatment with VB [b]	6 (3.6)	2 (2.3)	4 (5.1)	.35

[a] Continues variables expressed as mean ± standard deviation; all remaining continuous variables are expressed as median (IQR)
[b] Data displayed as n (%)
VB = vacuum bell

VB therapy for female patients

There was no difference in outcomes between female and male patients (P=.78). Female patients (n=28) started treatment at a younger age compared to male patients (11.5y (IQR 10.0-13.8) versus 15.0y (IQR 14.0-16.0), P<.001). The treatment duration (months) was equivalent to male patients (20.5 (IQR 13.0-43.5) versus 23.0 (IQR 12.0-32.0), P=.49), which resulted in a younger age for female patients at the end of treatment (13.0 (IQR 11.5-15.0) versus 17.0 (IQR 16.0-18.0), P<.001). Breast growth caused 39.3% (11/28) of female patients to quit treatment. Eight of them were treated unsuccessfully and three were lost to follow-up.

Similar to the overall patient group, there was no relationship between type of VB and treatment outcomes in female patients (P=.15). Only 9.1% (1/11) of female patients who stopped treatment because of breast growth was wearing a woman type VB.

Prognostic factors

Both objective and subjective deeper PE were associated with a lower rate of success (resp. P=.02, P=.009), with a median sternal depression of 1.8 (IQR 1.5-2.5) versus 2.1 (IQR 1.9-2.5) centimeters in the successfully versus unsuccessfully treated group. Sternal depression was evaluated as mild/light in 44.2% of all patients treated successfully,

compared to 26.6% in the unsuccessfully treated patients. Conversely, severe sternal depression was noted in only 16.3% of the successfully treated patients, compared to 35.4% in the unsuccessfully treated group.

Chest wall flexibility was related to age (P=.008), but not to treatment duration (P=.97). Fewer successfully treated patients had a flexible chest wall (38.4%) compared to unsuccessfully treated patients (59.5%) (P=.007). Patients with a symptomatic PE (P=.02), especially with endurance problems (P=.009), had a lower chance of success. Clinical outcomes did not vary in comparing patients under the age of ten (n=12), eleven (n=20), and twelve (n=31) to those over the ages of ten, eleven and twelve (P=.72, P=.65, P=.23, respectively).

Previous treatment for pectus deformities (Ravitch surgery (n=3), Nuss procedure (n=2) or DCS-bracing (n=2)) had no influence on treatment outcomes (P=.59).

A higher percentage of successfully treated patients wore the VB overnight compared to those unsuccessfully treated (58.1%, 50/87 versus 30.4%, 24/78, P<.001). Nighttime use correlated with longer treatment duration (26.0 (IQR 18.0-38.0) versus 20.0 (IQR 12.0-26.8) months, P=.004).

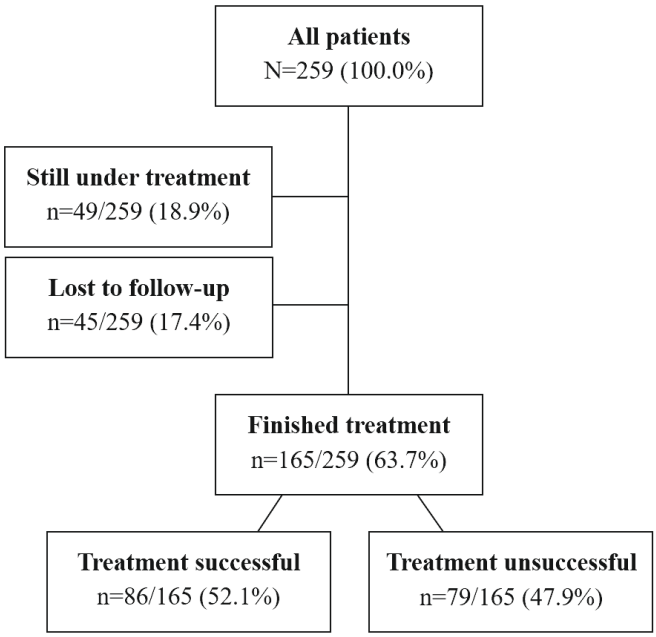


Figure 1 Flowchart showing treatment status and patient outcomes (n / N = number)

DISCUSSION

Of all patients finishing treatment, 52.1% finished treatment successfully. Factors contributing to a successful treatment were longer hours of daily VB use, overnight VB use and a prolonged treatment duration. A flexible thorax, deeper deformity and symptomatic PE were related to worse outcomes. Complications during treatment were minor and there were no long-term complications.

Success of VB treatment

Despite 52.1% of all patients finishing treatment achieving success, loss to follow-up was 17.4%. Median treatment time before being lost to follow-up was ten months (IQR 2.0-18.0), compared to 24.0 months (IQR 20.0-38.0) for successfully treated patients. This suggest those lost to follow-up may not have achieved favorable outcomes. If those patients are considered as treatment failures, the success rate would drop to 41.0% (86/210), which is comparable with the largest available study until now, which reported 43.6% (61/140).[7]

There is however, considerable variability in defining success among the literature. Some studies use a formula based on change in PE depth.[10, 11, 16] While useful for monitoring progress, this formula is based on a small reference group (n=30), and it does not capture the satisfaction and subjective assessment of the deformity by patients, parents and surgeons. Although objective measurement can guide treatment decisions, the final assessment of success should involve consultation with patients, parents and the surgeon, due to the significant subjective component of PE.

Lost to follow-up

Addressing the substantial percentage of patients lost to follow-up stands as the primary challenge in VB therapy. Our study had a loss to follow-up of 17.4%, comparable to other studies (3% to 36.1%).[7, 10, 11, 16-18]

Three important factors contribute to this. First, we lost contact with some patients nearing the end of treatment and switching to retainer mode, though they likely still use the VB for maintenance. Despite the positive impact of VB therapy, they are nevertheless categorized as lost to follow-up. Still, this group comprises only a small portion of drop-out, since most patient are lost to follow-up in an early stage. More importantly, the Amsterdam UMC covers PE treatment costs for patients under 18, which might reduce financial motivation and adherence to therapy. Additionally, once patients turn eighteen they must pay for the treatment themselves, which might also result in loss to follow-up.

A solution may be to call patients more often. Currently, patients are contacted by phone between the first and third month, and again at six months after initiating treatment. Their first physical check-up typically occurs after one year, although there is some variance between surgeons.

Daily time spend on VB therapy and treatment duration

Our results, which show that more daily hours of VB therapy lead to better results, are consistent with others, which showed better results with an average daily use of >6 hours,[16] >4 hours,[8] and >2 hours.[17] Additionally, two studies advocated wearing the VB >12 months.[10, 11]

In our opinion, based on these studies and our own results, more hours of daily wearing leads to better results, but we do not suggest a specific number of hours. The most effortless and comfortable way to add more hours is to wear the VB at night, despite described difficulties with VB dropping off or deflating. Although using the VB overnight was associated with better outcomes, treatment duration was a confounding factor since those also had longer treatment durations. Nonetheless, we strongly advise overnight use since it is the easiest way to add hours.

Long-term follow-up and complications

During our follow-up of more than four years recurrence occurred in 2.3% (2/86) of patients, which shows that results of VB therapy last. One of the patients with recurrence started VB therapy before the growth spurt. Similar to DCS-bracing, starting too early might cause recurrence during puberty.[19] The amount and type of complications were comparable to the literature.[14, 16] Pain, reported by 6.2% of patients, was associated with worse outcomes. We have not identified a cause of the pain.

VB therapy for patients awaiting the Nuss procedure

Some patients (15) opted for VB therapy while awaiting a Nuss procedure. This resulted in 26.7% (4/15) either finishing treatment successfully, or still being treated, without needing a subsequent Nuss procedure. Although this group is small, this suggests VB therapy during waiting periods may reduce the need for Nuss procedures.

Although there is a long waiting list at our Amsterdam Pectus Centre, other centers may not face this issue. In such cases, we recommend reserving VB therapy for patients with mild PE or those who strongly wish to avoid surgery.

VB therapy for female patients

The results of VB therapy in males and females are equivalent. Female patients start treatment earlier due to their earlier onset of puberty and focus on physical appearance. Breast development seems to be associated with early termination of treatment, causing 39.3% (11/28) of female patients to quit treatment, finishing treatment unsuccessfully or dropping out. This could also be a result of breast development which masks the PE, making further treatment unnecessary.

In all patient initially treated with a mini or small VB, clinical evaluation showed that the woman model VB did not fit either after the start of breast development. Currently there is no literature on the outcomes of the use of different models of VB in female patients. In our opinion, when breast development makes treatment of PE with a mini or small VB impossible, the woman model VB adds little to the treatment. Asymptomatic female patients often accept a moderate PE, since breasts masks the deformity.

Prognostic factors

Deeper sternal depth/depression and a flexible thorax correlated with worse outcomes. Age showed no outcome correlation, despite literature suggesting various upper age limits (twelve years,[13] eleven years[10, 18]). Interestingly, our study population was older (median age 15.0) compared to other studies (11.5-14.0).[8, 10, 16-18] Prada Arias et al. argued that young age leads to good results because of a flexible chest wall. In our study the relationship between a young age and a more flexible chest wall was confirmed.[16]

However, our finding that a flexible chest wall leads to worse outcomes contrasts with this and other studies, which often cite flexibility as a predictor of success.[10, 14] This discrepancy may be due to our subjective method for assessing thorax flexibility. Therefore we still believe that flexibility should be regarded as a positive prognostic factor, as supported by existing literature.

The prolonged treatment duration in our successfully treated patients (24.5 months, compared to 10.0-25.7 in the literature)[7, 8, 16] might be the key in explaining their good outcomes despite older age and a stiffer chest well, with the latter likely contributing to lasting results. Of note, not all studies mention exact treatment duration.[10] This heterogeneity makes comparison on treatment duration with other publications challenging.

Additionally, the follow-up periods in other studies are relatively short (average follow-up of 27.6, 12, 12, 18 and 40 months respectively,[7, 10, 13, 17, 18] or not mentioned[8]) compared to our median follow-up of 64.0 months. Shorter follow-up periods may miss

recurrences during or after the growth spurts in young patients, who were initially treated successfully in a short period of time due to their flexible chest wall.

Our results suggest that VB therapy can be successful and lasting in older patients with a stiff chest wall if treatment duration is long enough and patients remain motivated.

Two patients were treated with VB therapy after initial Nuss correction, but in both cases, treatment was unsuccessful. Despite only two cases, this suggests that further treatment after initial correction with a Nuss procedure should not be recommended.

Limitations

Some of the conclusions from our sub-analysis are based on results from a small number of patients. These conclusions need to be tested in larger populations to ensure they are reliable and generalizable.

Some variables had a high percentage of missing data, making multivariable logistic regression challenging. We recommend considering this approach for future studies.

Conclusions

VB therapy has a success rate of up to 52.1%, depending on how patients lost to follow-up are considered. Complications were minor. Prolonged treatment and overnight VB use to add extra treatment hours should be encouraged. Older patients with a stiff chest wall can be successfully treated with prolonged treatment. VB therapy is recommended while awaiting the Nuss procedure. Early treatment in female patients is preferred to avoid wearing the brace during breast growth.

REFERENCES

1 Brochhausen C, Turial S, Müller FK, et al. Pectus excavatum: history, hypotheses and treatment options. Interact Cardiovasc Thorac Surg. 2012;14:801-6.

2 Janssen N, Daemen JHT, van Polen EJ, et al. Pectus Excavatum: Consensus and Controversies in Clinical Practice. Ann Thorac Surg. 2023;116:191-9.

3 Ewert F, Syed J, Kern S, et al. Symptoms in Pectus Deformities: A Scoring System for Subjective Physical Complaints. Thorac Cardiovasc Surg. 2017;65:43-9.

4 Aly MR, Farina JM, Botros MM, et al. Minimally invasive repair of pectus excavatum in adults: a review article of presentation, workup, and surgical treatment. J Thorac Dis. 2023;15:5150-73.

5 Park HJ, Lee SY, Lee CS. Complications associated with the Nuss procedure: analysis of risk factors and suggested measures for prevention of complications. J Pediatr Surg. 2004;39:391-5.

6 Haecker FM. Evolution in the management of pectus excavatum in pediatric patients. Transl Pediatr. 2023;12:1450-3.

7 Haecker FM, Sesia S. Non-surgical treatment of pectus excavatum. J Vis Surg. 2016;2:63.

8 Lopez M, Patoir A, Costes F, et al. Preliminary study of efficacy of cup suction in the correction of typical pectus excavatum. J Pediatr Surg. 2016;51):183-7.

9 Scalise PN, Demehri FR. The management of pectus excavatum in pediatric patients: a narrative review. Transl Pediatr. 2023;12:208-20.

10 Obermeyer RJ, Cohen NS, Kelly RE Jr., et al. Nonoperative management of pectus excavatum with vacuum bell therapy: A single center study. J Pediatr Surg. 2018;53:1221-5.

11 Toselli L, Chinni E, Nazar-Peirano M, et al. Determinants of success associated with vacuum bell treatment of pectus excavatum. J Pediatr Surg. 2022;57:550-4.

12 Gao Y, Li JH, Yu JG, et al. Noninvasive treatment of pectus excavatum with a vacuum bell combined with a three-dimensional scanner. Pediatr Surg Int. 2020;36:1205-11.

13 Yi E, Lee K, Jung Y, et al. Finding suitable candidates for vacuum bell therapy in pectus excavatum patients. Sci Rep. 2021;11:22787.

14 Patel AJ, Hunt I. Is vacuum bell therapy effective in the correction of pectus excavatum? Interact Cardiovasc Thorac Surg. 2019;29:287–90.

15 Sesia SB, Hradetzky D, Haecker FM. Monitoring the effectiveness of the vacuum bell during pectus excavatum treatment: Technical innovation. J Pediatr Surg. 2018;53:411-7.

16 Prada Arias M, Gómez Veiras J, Rodríguez Iglesias P, et al. Treatment of pectus excavatum with vacuum bell during puberty. Cir Pediatr. 2023;36:116-21.

17 St-Louis E, Miao J, Emil S, et al. Vacuum bell treatment of pectus excavatum: An early North American experience. J Pediatr Surg. 2019;54:194-9.

18 Lei W, Shao M, Hu Y, et al. Vacuum bell therapy for pectus excavatum: a retrospective study. BMC pediatr. 2024;24:173.

19 Van Braak H, de Beer SA, Zwaveling S, et al. Ravitch Surgery or Dynamic Compression Bracing for Pectus Carinatum: A Retrospective Cohort Study. Ann Thorac Surg. 2024;117:144-50.

Chapter 7

Intercostal nerve cryoablation or epidural analgesia for multimodal pain management after the Nuss procedure: a cohort study.

Hendrik van Braak, Sjoerd A. de Beer, Justin R. de Jong, Markus F. Stevens, Gijsbert D. Musters, Sander Zwaveling, Matthijs W.N. Oomen, Wendeline J. van der Made, Egbert Krug, L.W. Ernest van Heurn

European Journal of Pediatric Surgery. 2024 Dec 34:488-492.

ABSTRACT

Background

Nuss procedure for pectus excavatum is a minimally invasive, but painful procedure. Recently, intercostal nerve cryoablation has been introduced as a pain management technique.

Materials and methods

In this cohort study, we compared the efficacy of multimodal pain management strategies in children undergoing a Nuss procedure. The effectiveness of intercostal nerve cryoablation combined with patient-controlled systemic opioid analgesia (PCA) was compared to continuous epidural analgesia (CEA) combined with PCA. The study was conducted between January 2019 and July 2022. Primary outcome was length of stay (LOS), secondary outcomes were operation room time, postoperative pain, opioid consumption and gabapentin use.

Results

Sixty-six consecutive patients were included, 33 patients in each group. The cryoablation group exhibited lower Numeric Rating Scale (NRS) pain scores on postoperative day one and two (P=.002, P=.001) and a shorter LOS (three versus six days (P<.001)). Cryoablation resulted in less patients requiring opioids at discharge (30.3% versus 97.0% (P<.001)) and one week after surgery (6.1% versus 45.4% (P<.001)). In the CEA group gabapentin use was more prevalent (78.8% versus 18.2%, P<.001) and the operation room time was shorter (119.4 versus 135.0 minutes (P<.010)). No neuropathic pain was reported.

Conclusions

Intercostal nerve cryoablation is a superior analgesic method compared to CEA, with reduced LOS, opioid use and NRS pain scores. The prophylactic use of gabapentin is redundant.

INTRODUCTION

Pectus excavatum (PE) is a common chest wall deformity that results from sternal displacement due to rib cartilage overgrowth. The incidence is estimated at 1 in 40-400 individuals.[1] Conventional treatment for PE is minimally invasive repair of pectus excavatum (MIRPE), known as the Nuss procedure.[2]

This procedure causes significant postoperative pain, often resulting in an extended length of hospital stay (LOS) and increased opioid use. In most hospitals, thoracic continuous epidural analgesia (CEA) is the primary method of pain management after the Nuss procedure.[3] Because increased infection risk limits its use to three days, CEA is often supplemented with patient-controlled systemic opioid analgesia (PCA).

Thoracoscopic intercostal nerve cryoablation offers an alternative postoperative pain management approach. Alone or combined with the standard of care in multimodal pain protocols, it reduces postoperative opioid use and LOS.[2, 4, 5] However, uncertainties remain regarding long-term complications, particularly neuropathic pain (for which prophylactic gabapentin is used). Although the abundance of studies, pain scores in the only randomized controlled trial did not differ between cryoanalgesia and CEA.[2] Furthermore, most studies do not report pain scores or the incidence of opioid medication at discharge or later.

We share our experience with intercostal nerve cryoablation and CEA in the Amsterdam Pectus Center, with special focus on postoperative pain scores and the need for prophylactic gabapentin use.

PATIENTS AND METHODS

Ethics

Given the nature of this research, not interfering with the treatment of patients, an official waiver of ethical approval (W20_235 # 20.271) was granted by the Medical Research Ethics Committee (METC) of the Amsterdam UMC, Amsterdam, the Netherlands (Chairperson MSc. O. Harlaar) on 28 May 2020. Informed consent was obtained from all individual patients (or their parents) included in this study.

Study population and design

We performed a cohort study of patients (<18 years) undergoing a Nuss procedure at the Amsterdam Pectus Center and Maastricht University Medical Center between January 2019 and July 2022.

The prospectively included cryoablation cohort consisted of the first patients undergoing the Nuss procedure during our trial period with cryoablation and PCA. The retrospectively included CEA cohort consisted of the last patients undergoing the Nuss procedure with CEA and PCA. Both evenly sized cohorts consisted of consecutive patients. No case-matching was performed. The number of patient undergoing the Nuss procedure during the study period determined the sample size.

Patients received CEA combined with PCA for three/four days, or intra-operative bilateral thoracoscopic intercostal nerve cryoablation with PCA. If necessary, additional pain treatment was administered according to our pain protocol. No other intraoperative nerve blocks, preoperative analgesics or anxiolytics were given.

Data extraction

Patient and treatment characteristics were retrospectively obtained from patient records. This included analysis of operative reports, radiology reports, and patient charts to gather information on patient demographics, primary treatment characteristics, operative technique, hospital stay, and outpatient follow-up.

Numeric Rating Scale (NRS) pain scores (average score of up to three daily NRS pain scores) were recorded on day zero (day of surgery), one day post-surgery and two days post-surgery. Opioid prescriptions were documented at discharge. One week after discharge, patients were contacted via telephone to gather information on opioid use. After six weeks, the patients visited the outpatient clinic for an evaluation of current medication and opioid use. Minimum follow-up period was twelve months.

Outcomes

Primary outcome was LOS. Secondary outcomes were opioid consumption, NRS pain scores, use of PCA, operation room time, and gabapentin use.

Statistical analysis

Descriptive measurements were utilized to characterize the study population. The Shapiro-Wilk test was employed to determine parametricity of continuous variables. Nominal or categorical data were analyzed using Fisher's exact test or the chi-square test. Continuous variables were analyzed using the Mann-Whitney U test or the Independent Sample T-Test. Statistical significance was set at a probability value of less than 0.05. Data were analyzed using IBM SPSS Statistics version 28.0.

RESULTS

Patient characteristics

All patients undergoing the Nuss procedure were examined for eligibility, all (66) patients in the study period were confirmed to be eligible and included in the study. None of the patients was lost to follow-up. Thirty-three patients were included in each cohort. Of all patients, 84.8% were male (56/66) and median age was 16.0 years. Most patients received one Nuss bar (84.8%, 56/66). Median follow-up was 35.2 (IQR 17.0 to 44.9) months. No readmissions were reported. Baseline characteristics are described in Table 1, perioperative and postoperative characteristics and outcomes are described in Table 2.

Operation room time, pain management and opioid consumption

Mean operation room time was 15.6 (95% CI -2.2 to -29.0) minutes shorter for the CEA group, P<.010) compared to the cryoablation group. There was no significant difference in operation room time between the Nuss procedure with one or two bars (127.0 versus 132.0, P=.365).

NRS pain scores at day one and two were lower in the cryoablation group (respectively -1.3 (95% CI -2.2 to -.4), P=.002 and -2.0 (95% CI -.5 to -3.0), P=.001). Use of gabapentin (78.8%, 26/33 versus 18.2%, 6/33) was higher in the CEA group (P<.001).

Differences in opioid use at discharge, after one and after six weeks are visualized in Figure 2. Fewer patients used opioids at discharge (10/33, 30.3% versus 32/33, 97.0%) (P<0.001)) and one week after surgery (2/33, 6.1% versus 15/33, 45.4% (P<.001)) in the cryoablation group. After six weeks two patients still used opioids, one in each group. The use of PCA after surgery was shorter in the cryoablation group (1.6 days versus 4.1 days, P<.001).

LOS

Median LOS was three days shorter in the cryoablation group (Figure 1). In the cryoablation group, seven patients stayed for five days or longer (18 in the CEA group). In two cases the prolonged stay was related to the cryoablation procedure (large bilateral pneumonia (n=1), pain due to unilateral cryoablation (n=1)). Other reasons for prolonged hospital stay were social problems (n=2), opioid related gastrointestinal problems (n=2) and persistent pain (n=1).

DISCUSSION

Intercostal nerve cryoablation combined with PCA as multimodal pain management for Nuss procedure leads to clinically relevant reduction in pain levels compared to the combination of CEA with PCA. This leads to reduced LOS and a reduction in the use of opioids postoperatively, at discharge and one week after discharge.

Table 1. Patient characteristics

	All	**CEA**	**Intercostal nerve cryoablation**
	(N=66)	*(n=33)*	*(n=33)*
Sex [b]			
Male	56 (84.8)	27 (81.8)	29 (87.9)
Female	10 (15.2)	6 (18.2)	4 (12.1)
Age [a]	16.0 (15.0-17.0)	16.0 (14.0-16.0)	16.0 (16.0-17.0)
Follow-up (months) [a]	35.2 (17.0-44.9)	44.8 (42.3-47.4)	17.0 (15.4-32.9)
Severity of pectus excavatum [b]			
Light	11 (16.6)	3 (9.1)	8 (24.2)
Moderate	45 (68.2)	23 (69.7)	22 (66.7)
Severe	10 (15.2)	7 (21.2)	3 (9.1)
Nuss bar(s) [b]			
One	56 (84.8)	24 (72.7)	32 (97.0)
Two	10 (15.2)	9 (27.3)	1 (3.0)

[a] Nonparametric continuous variables, expressed as median (IQR)
[b] Data displayed as n (%)
CEA = continuous epidural analgesia, N/n = number

Table 2. Duration of surgery, pain scores, length of hospital stay, opioid consumption and complications

	CEA	**Intercostal nerve cryoablation**	**P-value**
	(n=33)	**(n=33)**	
Operation room time (min.) [a]	119.4 ± 22.7	135.0 ± 31.3	.010
Postoperative VAS score			
Day 0 [a]	3.1 ± 1.9	2.4 ± 1.8	.133
Day 1 [a]	4.2 ± 1.9	2.9 ± 1.7	.002
Day 2	4.0 (2.5-5.0)	2.0 (2.0-3.0)	.001
Stop of PCA (days after surgery) [a]	4.1 ± 1.9	1.6 ± .9	<.001
LOS (days)	6.0 (5.0-8.0)	3.0 (2.0-4.0)	<.001

Table 2. Duration of surgery, pain scores, length of hospital stay, opioid consumption and complications *Continued*.

	CEA (n=33)	Intercostal nerve cryoablation (n=33)	P-value
Opioid use [b]			
At discharge	32 (97.0)	10 (30.3)	<.001
At discharge + 1 week	15 (45.5)	2 (6.1)	<.001
At discharge + 6 weeks	1 (3.0)	1 (3.0)	1.000
Gabapentin use [b]	26 (78.8)	6 (18.2)	<.001

[a] Parametric variables; expressed as mean ± standard deviation; all remaining continuous variables are nonparametric and expressed as median (IQR).
[b] Data displayed as n (%)
CEA = continuous epidural analgesia, LOS = length of hospital stay, N/n = number, min. = minutes, PCA = patient-controlled systemic opioid analgesia, P-value = probability value, VAS = visual analogue scale

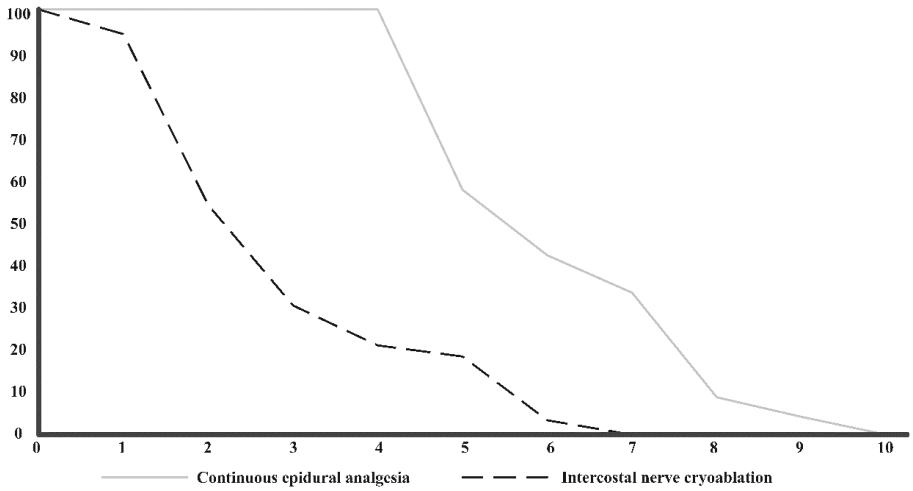


Figure 1 Comparison of length of hospital stay in both treatment groups (Y-axis: percentage of patients still hospitalized, x-axis: days)

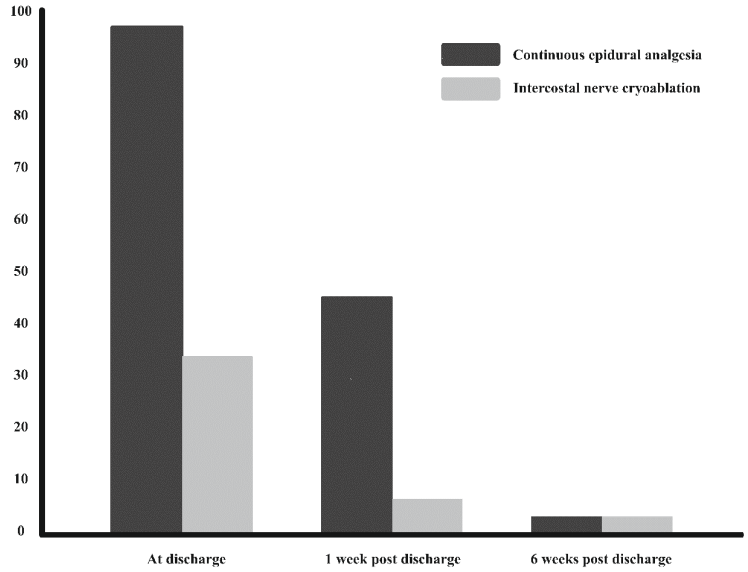


Figure 2 Comparison of opioid use in both treatment groups (Y-axis: percentage of patients using opioids)

Operation room time

This study shows a comparable operation room time between CEA and cryoablation, similar to the only other study reporting on this matter.[3] Since epidural placement is essential in the Nuss procedure, we consider operation room time more relevant than surgery time when comparing both treatments. Our findings put the argument that cryoablation is time-intensive, and consequently more expensive compared to CEA, into perspective.

Postoperative pain

Directly postoperative (day zero) the NRS pain scores were similar between both groups, which is consistent with current literature.[2, 6, 7] Only one study demonstrated an advantage of cryoablation.[8] This was likely the effect of perioperatively titrated opioids and anesthetics. We reported statistically significant and clinically relevant differences in NRS pain scores on day one and two between the groups, which has not yet been described before.

Opioid use

Postoperative opioid use is defined in different ways. Most studies use total oral morphine milligram equivalents (MME) to assess differences in opioid use. We measured the percentage of patients using opioids or PCA at specific time points. Holguin et al.

used both methods and obtained similar results as ours (discontinuation of PCA after one day versus after four days), also showing a difference in percentage of patients requiring narcotics on discharge (100% versus 54.8%).[5] We achieved even better results at discharge, and further demonstrate that opioid usage after one week remains lower in the cryoablation group. This is a promising result, which has not yet been described before.

LOS

The LOS was three days shorter in the cryoablation group, which is similar to the literature (LOS varying from -2 to -3 days).[3-5, 8]

Since cryoablation was implemented after years of providing extensive multimodal pain therapy, requiring long hospitalization, patients in the cryoablation cohort might have been hospitalized for too long. An analgesic regimen that further reduces opioid use may reduce LOS even further.[3] This suggests the possibility of same-day discharge, as supported by promising results from a pilot study.[9]

Neuropathic pain and gabapentin use

We reported no patients with neuropathic pain, which is consistent with current literature.[10] Despite the low incidence of neuropathic pain in adolescents, standard prescription of prophylactic gabapentin and/or muscle relaxants during hospitalization and/or after discharge, is described in two studies.[3, 6] Others used gabapentin only on indication.[2, 5] However, all studies reported a low incidence of neuropathic pain. In our population gabapentin (300 mg, twice daily for five days) was prescribed postoperatively in all patients weighing >40 kilograms in the CEA group and in six patients in the cryoablation group.

Limitations

The characteristics of the study and the small sample size limits the generalizability of the results. However, there is a large difference between both groups, both in use of analgesics and LOS.

Conclusion

We conclude that intercostal nerve cryoablation is a superior analgesic method compared to CEA, with reduced LOS, opioid usage and NRS pain scores. We recommend against the routine use of gabapentin and leave its use for specific neuropathic pain.

REFERENCES

1 Biavati M, Kozlitina J, Alder AC, et al. Prevalence of pectus excavatum in an adult population-based cohort estimated from radiographic indices of chest wall shape. PloS One 2020;15:e0232575.

2 Graves CE, Moyer J, Zobel MJ, et al. Intraoperative intercostal nerve cryoablation during the Nuss procedure reduces length of stay and opioid requirement: A randomized clinical trial. J Pediatr Surg 2019;54:2250-6.

3 Sun RC, Mehl SC, Anbarasu CR, et al. Intercostal cryoablation during Nuss procedure: A large volume single surgeon's experience and outcomes. J Pediatr Surg 2021;56:2229-34.

4 Daemen JHT, de Loos ER, Vissers YLJ, et al. Intercostal nerve cryoablation versus thoracic epidural for postoperative analgesia following pectus excavatum repair: a systematic review and meta-analysis. Interact Cardiovasc Thorac Surg 2020;31:486-98.

5 Perez Holguin RA, DeAngelo N, Sinha A, Shen C, Tsai AY. Cost and outcomes of intercostal nerve cryoablation versus thoracic epidural following the Nuss procedure. J Pediatr Surg 2023;58:608-12.

6 Keller BA, Kabagambe SK, Becker JC, et al. Intercostal nerve cryoablation versus thoracic epidural catheters for postoperative analgesia following pectus excavatum repair: Preliminary outcomes in twenty-six cryoablation patients. J of Pediatr Surg 2016;51:2033-8.

7 Harbaugh CM, Johnson KN, Kein CE, et al. Comparing outcomes with thoracic epidural and intercostal nerve cryoablation after Nuss procedure. J Surg Res 2018;231:217-23.

8 Aiken TJ, Stahl CC, Lemaster D, et al. Intercostal nerve cryoablation is associated with lower hospital cost during minimally invasive Nuss procedure for pectus excavatum. J Pediatr Surg 2021;56:1841-5.

9 Akinboro S, John R, Reyna T, et al. A pilot study of multi-modal pain management for same-day discharge after minimally invasive repair of pectus excavatum (Nuss procedure) in children. Pediatr Surg Int 2023;39:159.

10 Zobel MJ, Ewbank C, Mora R, et al. The incidence of neuropathic pain after intercostal cryoablation during the Nuss procedure. Pediatr Surg Int 2020;36:317-24.

Chapter 8

Pleural effusion after Nuss procedure: a case series

Hendrik van Braak, Suzanne W.J. Terheggen-Lagro, Joël Israels, Joost van Schuppen, Eline E. Deurloo, Justin R. de Jong

Journal of Pediatric Surgery Case Reports. 2025 Aug 121:103085.

ABSTRACT

Introduction
Pleural effusion is an underreported and poorly understood complication of the Nuss procedure.

Cases presentation
Case 1 A 16-year-old female presented five weeks postoperatively with dyspnea (CRP 258mg/L), left-sided pleural and pericardial effusion. Drainage and treatment with amoxicillin/clavulanic acid, gentamicin, diuretics, ibuprofen, and prednisolone led to improvement. After steroid discontinuation, effusion recurred despite treatment with amoxicillin/clavulanic acid and gentamicin. Cultures remained negative. She required thoracic drainage and received oral clindamycin, later switched to ciprofloxacin, with eventual recovery.

Case 2 A 17-year-old male presented with right-sided chest pain and pleural effusion two weeks after a complicated Nuss procedure (due to extensive adhesiolysis for prior pneumonia-related lung adhesions. Despite antibiotics, symptoms progressed, and he was admitted (CRP 240mg/L). Chest drainage yielded 1300mL clear fluid; cultures were negative. Recurrent effusion eventually required video-assisted thoracoscopic surgery. He was treated with amoxicillin/clavulanic acid after surgery.

Case 3 A 14-year-old female developed fever one week postoperatively (CRP 123mg/L), with no abnormalities on imaging. Three weeks later, she developed right-sided effusion; 600mL was drained, and she received ceftriaxone and clindamycin. Recurrence occurred within two weeks; oral prednisolone was added, resulting in rapid improvement. She was discharged on cotrimoxazole and tapered steroids. Two months later, imaging for persistent fatigue revealed Nuss bar dislocation and pneumothorax, requiring bar repositioning.

Conclusion
Pleural effusion after the Nuss procedure can be caused by a reactive inflammatory response, triggered by pleural or mechanical irritation. If an infection and bar displacement are ruled out, a course of corticosteroid therapy may be effective.

INTRODUCTION

The Nuss procedure is currently the preferred treatment for pectus excavatum (PE). While outcomes are good, a range of complications has been reported in the literature.[1, 2]

One such complication, pleural effusion, has been described in up to 10.2% of patients postoperatively,[3, 4] yet its etiology remains unclear and likely multifactorial, with proposed contributors including multiple bar placement, sternal elevation, or metal allergy.[5-9] Although several treatment strategies have been described, including drainage, saline irrigation, corticosteroids, and non-steroidal anti-inflammatory drugs (NSAID's),[5, 8, 9] correctly diagnosing pleural effusion as primary problem, and distinguishing it from pneumonia, is clinically challenging. This is due to overlapping features such as fever, chest pain, dyspnea, and elevated inflammatory markers, which can lead to misdiagnosis, delayed care, and unnecessary treatments.

We report a case series of patients who developed pleural effusion mimicking complex pneumonia following the Nuss procedure and propose a stepwise approach for early identification and management. This manuscript was prepared following the CARE guidelines.

CASES PRESENTATION

All cases described in this series were managed at the Amsterdam Pectus Center, a high-volume referral center within the Amsterdam University Medical Center. With over 100 Nuss procedures performed annually, our institution has extensive experience in the surgical treatment of pectus excavatum. Over the past 15 years, the incidence of late-onset pleural effusion following the Nuss procedure has been 2.4%. The following three cases illustrate the diagnostic and therapeutic challenges associated with this postoperative complication.

Case 1

The patient, a sixteen-year-old female, presented with dyspnea and general discomfort nearly five weeks after an uncomplicated Nuss procedure (epidural analgesia, one Nuss bar, stabilizer on the right side, no use of crane technique). Surgical sites showed no signs of inflammation. C-reactive protein (CRP) was 258 mg/L. A CT-scan was performed, revealing left-sided pleural effusion, pericardial effusion, and a fluid collection between the liver and heart, raising suspicion of post-pericardiotomy syndrome (PPS). Fluid was drained, and the patient was treated with amoxicillin/clavulanic acid, gentamicin, diuretics, ibuprofen, and intravenous prednisolone. Clinical improvement was observed,

with CRP decreasing to 19 mg/L and the patient was discharged after one week with a prescription for oral amoxicillin/clavulanic acid.

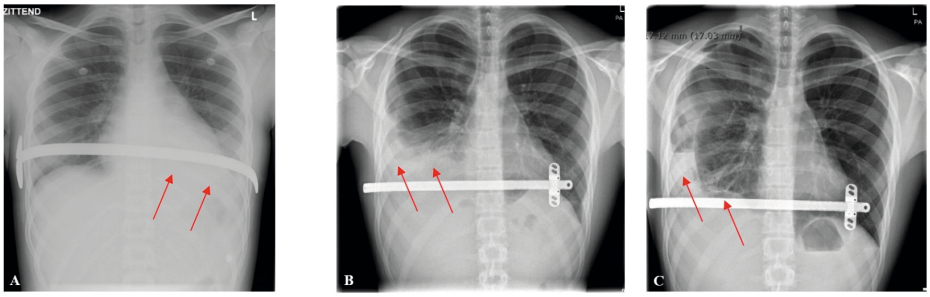


Figure 1 X-ray of the chest of patient one at second admission, seven weeks after surgery, with arrows indicating left-sided pleural effusion. The heart is also slightly enlarged (A). X-ray of the chest of patient two at first presentation, two weeks after surgery, with arrows indicating right-sided pleural effusion (B). X-ray of the chest of patient two at routine follow-up before second admission, four days after discharge, with arrows indicating right-sided pleural effusion and pneumothorax (C).

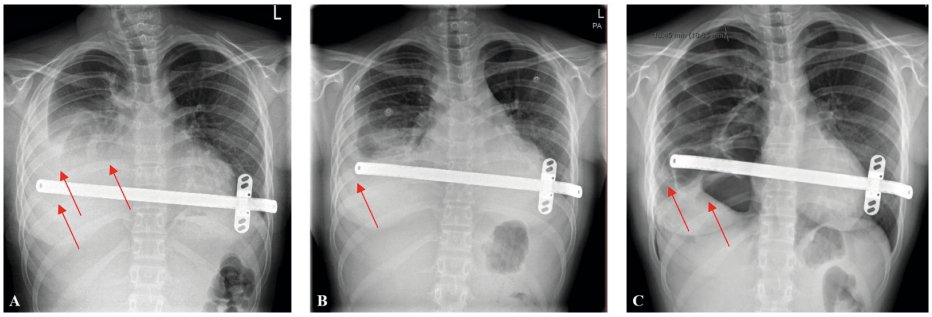


Figure 2 X-ray of the chest of patient three at first admission, two weeks after first presentation, with arrows indication pleural effusion and consolidation of the right lower and middle lobes with air bronchogram (A). X-ray of the chest of patient three at second admission, two weeks after discharge, showing atelectasis on both sides, with arrows indicating persistent pleural effusion (B). X-ray of the chest of patient three at third admission, two months after discharge from second admission, showing increased volume loss of the right lung with progression of bullae formation (C).

Two weeks later, the patient was readmitted with recurrent symptoms of dyspnea and general discomfort. CRP was 26 mg/L and imaging still showed left-sided pleural and pericardial effusion and atelectasis of the left lung (Figure 1A). Despite initiation of intravenous amoxicillin/clavulanic acid and gentamicin, CRP quickly rose to 210 mg/L. A thoracic drain was placed after three days, leading to symptom relief and a reduction in inflammatory parameters. Bacterial cultures of pleural fluids remained negative. The

patient was discharged after two weeks with oral clindamycin, which was later switched to amoxicillin/clavulanic acid due to an allergic reaction.

One week after discharge, the patient was again readmitted with dyspnea, general discomfort and a CRP of 154mg/L under antibiotic treatment. Gentamicin was reintroduced, leading to symptom improvement. Allergy testing for metals in the Nuss bar was negative, and a bone scan ruled out osteomyelitis. The patient was discharged after two weeks with oral ciprofloxacin, after which symptoms fully resolved.

Case 2

The patient, a seventeen-year-old male, presented with right-sided thoracic pain two weeks after a complicated Nuss procedure (cryoanalgesia, one Nuss bar, stabilizer on the left side, no use of crane technique). Intraoperatively, adhesions of the right lung to the thoracic wall were encountered during thoracoscopy, likely the result of childhood pneumonia. These adhesions required extensive adhesiolysis, significantly prolonging the duration of the surgery.

Upon presentation, surgical sites showed no signs of infection. CRP was 69 mg/L. A chest X-ray (Figure 1B) revealed a right-sided pleural effusion. The patient was initially managed on an outpatient basis with oral amoxicillin/clavulanic acid; however, the pain worsened. After one week the patient was admitted to the hospital with pain, fever, a CRP of 240 mg/L and progressive pleural effusion. A chest drain was placed, aspirating 1300 mL of clear fluid, and oral clindamycin was started. Cultures from the aspirated fluid remained negative. The drain was removed after two days, and the patient was discharged one day after drain removal. Four days later, the patient was readmitted due to an increasing pleural effusion, identified on routine follow-up chest X-ray (Figure 1C), without symptoms. A CT scan revealed a right-sided pleural effusion with multiple air collections within the pleural space, and possible consolidations, raising suspicion of an empyema. Intravenous amoxicillin/clavulanic acid was started and a Video-Assisted Thoracoscopic Surgery (VATS) was performed, revealing no pus but only fluid and residual hematomas. Additionally, intraoperative findings confirmed right lung adhesions to the chest wall. The chest drain was removed two days after the surgery, and amoxicillin/clavulanic acid was switched to oral administration. The patient was discharged after a week.

Case 3

The patient, a fourteen-year-old female, presented one week after an uncomplicated Nuss procedure (cryoanalgesia, one Nuss bar, stabilizer on the right side, no use of crane technique), with fever and a CRP of 123 mg/L. Surgical sites showed no signs of inflammation. An X-ray of the chest showed no abnormalities, and symptoms were attributed to influenza. Three weeks postoperatively, the patient returned with chest pain. The X-ray of the chest (Figure 2A) showed a consolidation with right-sided pleural effusion. A chest drain was placed, aspirating 600 mL of slightly cloudy fluid. Cultures from the aspirated fluid remained negative. The patient was treated with ceftriaxone and clindamycin. The drain was removed after three days, and after a hospital stay of eight days, she was discharged with a four-week course of oral amoxicillin/clavulanic acid and amoxicillin.

However, within two weeks after the first hospital admission, the patient presented with pain and fever. An X-ray of the chest revealed a new right-sided pleural effusion (Figure 2B). After two days of intravenous ceftriaxone and clindamycin, oral prednisolone was added, leading to rapid clinical improvement. The patient was discharged three days later with a three-week course of oral cotrimoxazole and prednisolone, which were tapered over the next five weeks.

The patient returned to the emergency department two months later with persistent fatigue. Infection parameters were not elevated. An X-ray of the chest (Figure 2C) revealed pneumothorax of the right lung, prompting a CT scan, which identified a dislocated Nuss bar. This dislocation was not visible on the X-ray of the chest. Surgical correction of the dislocation was done, but the right lung remained collapsed despite drainage. Ultimately, a VATS was attempted but had to be converted to an open thoracotomy to perform adhesiolysis of the lung. The patient required prolonged hospitalization but was eventually discharged after one month. This prolonged course and multiple readmissions had a large impact on this patient and her family.

DISCUSSION

Pleural effusion after the Nuss procedure is classified as early postoperative when detected before hospital discharge and late postoperative when identified thereafter. Early effusions, reported in up to 56.7 % of patients, are usually self-limiting and only occasionally require drainage.[10-12] Symptomatic pleural effusion requiring treatment occurs less frequent, with a reported incidence of 1.1-7.1%,[5, 8, 9, 11, 12] and typically presents several weeks after surgery (Table 1).[5] These late-onset effusions constitute a far more complex clinical entity.

Proposed risk factors for pleural effusion include multiple, crossed bars, possibly due to an increased metal load, metal allergy, and use of sternal elevation techniques such as the crane method.[5, 7-9] These factors likely share a common mechanism: increased pleural irritation, which may trigger inflammation leading to pleural effusion. The finding of a dislocated bar in case three supports this hypothesis. Cryoanalgesia may contribute similarly, as it causes local inflammation and involves substantial tissue handling.[13] Both case two and three received cryoanalgesia, warranting further investigation into its potential role in pleural effusion.

Although pleural effusion is easily seen on chest X-ray, distinguishing isolated late pleural effusion from complicated pneumonia is challenging as symptoms overlap (fever, pain, dyspnea, elevated inflammatory markers). Retrospective review by pediatric pulmonologist and radiologist confirmed that imaging, history, and labs cannot reliably differentiate sterile effusion from pneumonia, leading to potential treatment delays and unnecessary interventions. This dilemma was evident in all three cases, where effusion persisted despite antibiotics, suggesting an inflammatory rather than infectious cause. Two patients improved with corticosteroids, supporting the hypothesis of a hyperinflammatory response from pleural manipulation rather than infection. In addition to X-rays, ultrasound can reveal septations or possible bar involvement, raising suspicion for empyema or bar-related infection, but these findings are not definitive since longstanding, sterile effusions may also present with septations. Therefore, pleural fluid aspiration and culture remain essential as the diagnostic gold standard.

Currently, several treatment options for late pleural effusion after the Nuss procedure have been reported, although evidence remains limited (Table 1).[3, 5, 8-10, 12] Initially, however, literature was scarce. Because symptoms and imaging closely resembled pneumonia, we initially treated all effusions as complicated pneumonias with antibiotics and drainage. With current knowledge, we recognize that some effusions are reactive rather than infectious. Since distinguishing these entities is challenging and missing pneumonia can have serious consequences, we recommend starting antibiotics until infection is excluded. If cultures are negative or no improvement occurs within 72 hours, corticosteroids can be considered. Drainage remains useful for symptom relief and lung expansion. Based on our findings, we propose standardized assessment for all late effusions (Figure 3) and have advanced follow-up chest X-rays from six to three weeks postoperatively for earlier detection.

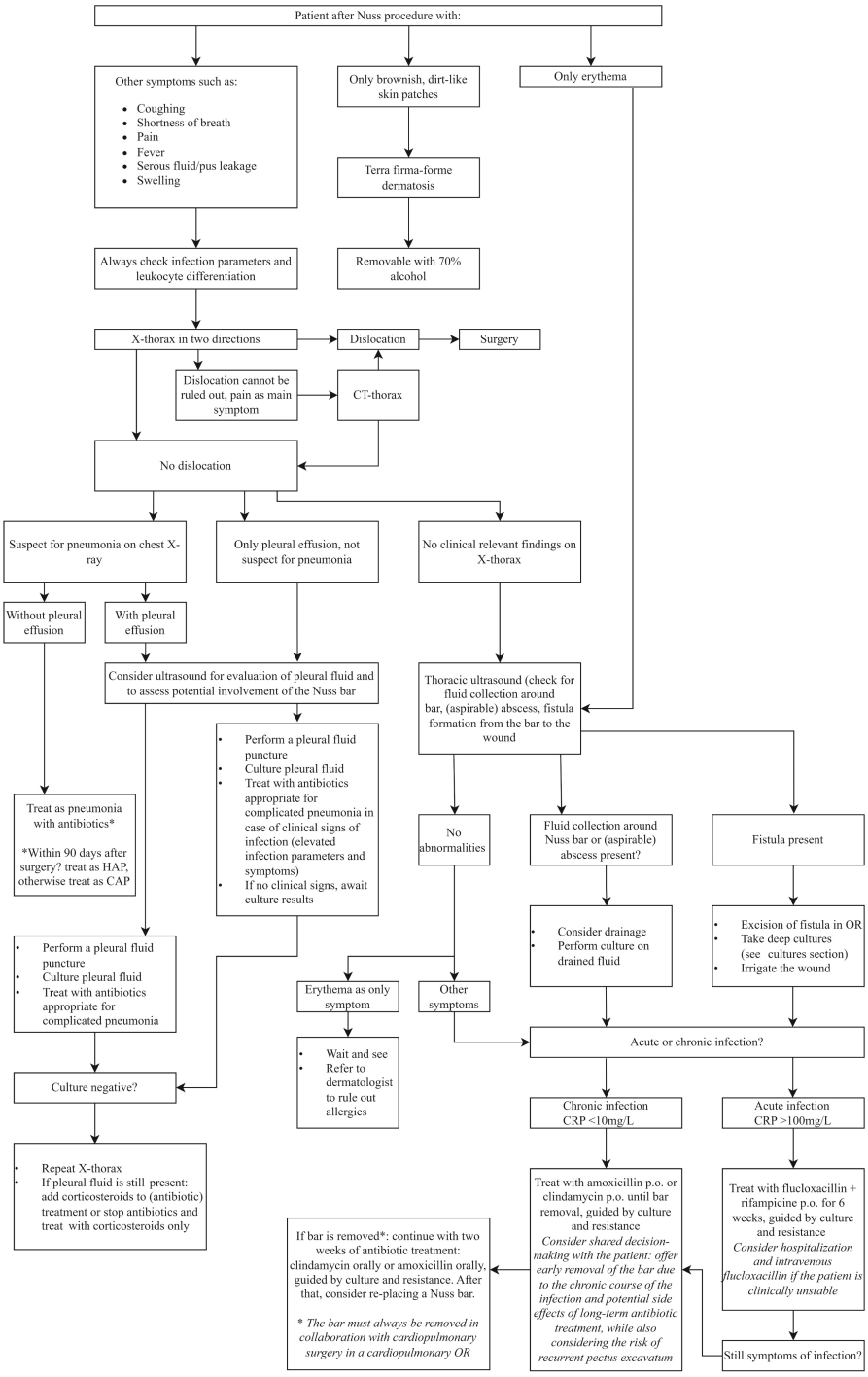


Figure 3 Protocol for managing postoperative Nuss bar infections (CAP = Community Acquired Pneumonia, CRP = C-Reactive Protein, HAP = Hospital Acquired Pneumonia)

Table 1 Summary of reported treatment strategies for pleural effusion following the Nuss procedure

Early postoperative pleural effusion [a]			
Author	Year	Incidence [b]	Treatment [b]
Croitoru et al.[10]	2002	56.7% (148/261)	Drainage (2.7% (4/148))
Castellani et al.[12]	2008	14.8% (26/176)	Drainage (23.1% (6/26))
Nuss et al.[3]	2016	0.9% (13/1463)	Drainage (100.0% (13/13))
Late postoperative pleural effusion [a]			
Author	Year	Incidence [b]	Treatment [b]
Castellani et al.[12]	2008	1.1% (2/176)	Repeated drainage and corticosteroids (100.0% (2/2))
Cheng et al.[5]	2014	2.6% (10/390)	Drainage (60.0% (6/10)) Indomethacin (90.0% (9/10)) Corticosteroids (30% (3/10))
Sayan et al.[8]	2021	2.2% (6/276)	Drainage (100.0% (6/6))
Oka et al.[9]	2024	7.1% (16/225)	Continuous pleural irrigation using saline and antibiotics (100.0% (16/16))

[a] Pleural effusion was classified as early postoperative when diagnosed before hospital discharge and as late postoperative when detected after discharge following the Nuss procedure
[b] Reported as % (n/N)

Besides ruling out bar infection, ruling out Nuss bar displacement is also essential. Case three illustrates this. Despite antibiotic and corticosteroid therapy, persistent symptoms were ultimately attributed to bar dislocation, visible only on CT scan and not on standard X-rays. In patients with unresolved or atypical symptoms after treatment, bar displacement should always be considered, and a CT scan should be performed.

One patient developed PPS alongside pleural effusion. PPS after the Nuss procedure occurs in about 2.4% of cases and is likely related to pericardial irritation during tunneling.[10, 14, 15] Corticosteroids and NSAIDs are effective but symptoms may recur, requiring prolonged treatment.[16] Since PPS likely results from pericardial irritation during tunneling, sternal elevation with the crane technique may reduce this risk by improving visualization and creating space, thereby minimizing the risk of pericardial traction or puncture. However, this introduces a paradox: while sternal elevation may prevent pericardial injury, it has also been identified as a potential risk factor for postoperative pleural effusion, reducing the risk of one complication while introducing another.[7]

As a retrospective case-series, data collection was dependent on available patient records, which may have introduced incomplete data retrieval. Furthermore, the retrospective nature of this study inherently restricts causal conclusions regarding the efficacy of different treatment strategies. While corticosteroids appears to be an effective treatment for pleural effusion based on our findings, only two cases provide direct evidence within our study. However, the safety and effectiveness of corticosteroids in similar cases are supported by the existing literature, making this a reasonable conclusion.[5,12] We aim to evaluate our new treatment protocol in the future.

CONCLUSION

Pleural effusion following the Nuss procedure presents a diagnostic and therapeutic challenge, as clinical symptoms and pleural effusion after the Nuss procedure can be caused by a reactive inflammatory response, triggered by pleural or mechanical irritation. If an infection and bar displacement are ruled out, a course of corticosteroid therapy may be effective.

REFERENCES

1. De Loos ER, Pennings AJ, van Roozendaal LM, et al. Nuss Procedure for Pectus Excavatum: A Comparison of Complications Between Young and Adult Patients. Ann Thorac Surg. 2021;112(3):905-11.
2. Akhtar M, Razick DI, Saeed A, et al. Complications and Outcomes of the Nuss Procedure in Adult Patients: A Systematic Review. Cureus. 2023;15(2):e35204.
3. Nuss D, Obermeyer RJ, Kelly RE. Nuss bar procedure: past, present and future. Ann Cardiothorac Surg. 2016;5(5):422-33.
4. Mehl SC, Sun RC, Anbarasu CR, et al. Association of Intercostal Nerve Cryoablation During Nuss Procedure With Complications and Costs. Ann Thorac Surg. 2023;116(4):803-9.
5. Cheng YL, Lin CT, Wang HB, Chang H. Pleural effusion complicating after Nuss procedure for pectus excavatum. Ann Thorac Cardiovasc Surg. 2014;20(1):6-11.
6. Rushing GD, Goretsky MJ, Gustin T, et al. When it is not an infection: metal allergy after the Nuss procedure for repair of pectus excavatum. J Pediatr Surg. 2007;42(1):93-7.
7. Anbarasu CR, Mehl SC, Sun RC, et al. Variations in Nuss Procedure Operative Techniques and Complications: A Retrospective Review. Eur J Pediatr Surg. 2022;32(4):357-62.
8. Sayan B, Bekiroglu N, Yuksel M. Pectus cross bars increase hospital readmission rates due to serous pleural effusion. Gen Thorac Cardiovasc Surg. 2022;70(4):352-8.
9. Oka N, Masai K, Okubo Y, et al. Clinical impact of multiple pectus bars on surgical outcomes following pectus excavatum repair. Interdiscip Cardiovasc Thorac Surg. 2024;39(4).
10. Croitoru DP, Kelly RE, Goretsky MJ, et al. Experience and modification update for the minimally invasive Nuss technique for pectus excavatum repair in 303 patients. J Pediatr Surg. 2002;37(3):437-45.
11. Goretsky MJ, McGuire MM. Complications associated with the minimally invasive repair of pectus excavatum. Semin Pediatr Surg. 2018;27(3):151-5.
12. Castellani C, Schalamon J, Saxena AK, Höellwarth ME. Early complications of the Nuss procedure for pectus excavatum: a prospective study. Pediatr Surg Int. 2008;24(6):659-66.
13. Ruz AD, Goyanes AG, Sierra CD, Javier Escribá Alepuz F, Pilar Argente Navarro M. P071 Risk assessment of intercostal cryoanalgesia in NUSS surgery: a case study on pleural effusion complications. Reg Anesth Pain Med. 2024;49:A284.
14. Yu PSY, Ng VWK, Lau RWH, Ng CSH. Massive pericardial effusion after Nuss procedure: to drain or not to drain? J Thorac Dis. 2018;10(1):E27-e30.
15. Nuss D, Croitoru DP, Kelly RE Jr., et al. Review and discussion of the complications of minimally invasive pectus excavatum repair. Eur J Pediatr Surg. 2002;12(4):230-4.
16. Muensterer OJ, Schenk DS, Praun M, Boehm R, Till H. Postpericardiotomy syndrome after minimally invasive pectus excavatum repair unresponsive to nonsteroidal anti-inflammatory treatment. Eur J Pediatr Surg. 2003;13(3):206-8.

Chapter 9

Nuss bar infections: risk factors and management strategies

Hendrik van Braak, Erika P.M. van Elzakker, Sjoerd A. de Beer, Matthijs W.N. Oomen, Sander Zwaveling, L.W. Ernest van Heurn, Justin R. de Jong

Submitted for publication

ABSTRACT

Background

This study aims to evaluate the incidence and characteristics of Nuss bar infections, identify surgical risk factors, and proposes an optimized management strategy.

Methods

We conducted a retrospective cohort study of all patients who underwent the Nuss procedure between 1999 and 2024 at the Amsterdam Pectus Center. Primary outcomes included the incidence and management of Nuss bar infections. Secondary outcomes included infection characteristics (early- versus late-onset, superficial versus deep), diagnostics, cultured bacteria, prognostic factors (sex, age, type of anesthesia, number of bars, bar dislocation, stabilizer position, use of sutures), impact on bar removal, and incidence of allergies and terra firma-forme dermatosis.

Results

Of 695 patients, 4.0% (28/695) developed Nuss bar infections, on average six months postoperative, presenting with erythema, pain and exudate. Over time, infections shifted from early- to late-onset (P=.03). Deep infections (71.4%, 20/28) more often required surgery (P=.007) whereas superficial infections were managed with antibiotics, involving a wide range of regimens. Bar dislocations (5.6%, 39/695) and stabilizer plate loosening (1.0%, 7/695) required reoperation and were associated with infections (P=.001). Operative time for bar removal was longer in infected patients (52.0 (32.0–68.0) versus 35.0 (26.0–49.0) minutes, P=.03). One case of cobalt allergy was identified. Terra firma-forme dermatosis (0.4%, 3/695) was successfully treated using alcohol wipes.

Conclusions

The incidence of Nuss bar infections remains low but requires structured, multidisciplinary management. Bar dislocation and plate loosening are key risk factors. Our protocol emphasizes precise diagnostics, consistent and tailored antibiotic therapy, and awareness of alternative diagnoses in atypical presentations.

INTRODUCTION

Pectus excavatum (PE) is a chest wall anomaly for which minimally invasive repair (MIRPE), first described by Nuss et al.,[1] is the most common treatment.[2] Despite favorable outcomes, postoperative infections remain a concern, with an incidence of 1.5%-9.4%.[3-15] For clean surgical wounds, a 1-5% infection rate is considered acceptable.[16] Affected patients often require extended antibiotic therapy and occasionally, additional surgical interventions, although the Nuss bar is typically retained.[4, 13, 17] The most comprehensive study on postoperative infections following MIRPE, including a protocol, was published in 2018.[4]

Since then, the surgical technique of the Nuss procedure has advanced significantly. In particular, the introduction of intercostal nerve cryoablation in 2019 has altered care.[18] We observed an increase in low-grade surgical postoperative infections alongside these advancements. We therefore reevaluated current protocols for prevention, diagnosis, and management of Nuss bar infections.

This study aims to evaluate the incidence and characteristics of Nuss bar infections, identify surgical risk factors, and propose a management strategy for postoperative infections.

PATIENTS AND METHODS

Study population and design

We performed a retrospective cohort study of patients undergoing MIRPE in the Amsterdam Pectus Center, Amsterdam University Medical Center, between March 1999 and April 2024. The Medical Ethics Review Committee of the Amsterdam Medical Center waived ethical approval. Informed consent was obtained from all patients (or their parents).

Data extraction

Data were retrospectively obtained from patient records. Infections were defined as cellulitis that required antibiotics, superficial infection with active drainage, or deep infections involving hardware, regardless of onset.[4] An infection was considered deep if ultrasound confirmed bar involvement, the bar or stabilizer plate became exposed, or imaging or surgery revealed direct connection between the infection and the bar (fistula/abscess). A 30-day threshold differentiated between early-onset and late-onset infections. Operative time for Nuss bar removal was defined as time between incision and wound closure. Rotation, lateral shift, flipping, or intrathoracic displacement are

all defined as Nuss bar dislocation. Stabilizer plate loosening is defined as fixation loss, allowing plate migration along the bar. Nuss bar allergy had to be confirmed by a dermatologist, through standardized patch testing.

Treatment protocol

Figure 1 shows our treatment protocol. Routine allergy testing is not part of our protocol. Until 2019, patients undergoing the Nuss procedure received continuous epidural analgesia combined with patient-controlled analgesia. Since then, the procedure has been performed using intercostal nerve cryoablation combined with patient-controlled analgesia for the first 24 hours after surgery.[18]

All patients receive intravenous cefazolin for 24 hours, starting 30 minutes before first incision (adolescents: 3x1000mg/day, children: 3x100–150mg/kg, maximum of 6gr/day). We follow the Dutch and international guidelines regarding surgical site infection prevention.[19, 20] Scrubbing is done twice with chlorhexidine/alcohol skin antiseptic solution.

We perform MIRPE using a modified version[21] of the original technique.[1] Typically one Nuss bar is used; although sometimes two bars are required. A single stabilizer plate on the left side of the bar, as medially as possible on the chest wall, secures the bar(s). In the early years, the bar (Biomet) was sutured to the stabilizer plate on the one side, and secured with a thoracoscopic placed pericostal, absorbable suture on the other side. We later adopted bars (MedXpert) that featured a stabilizer plate with screws, making sutures abundant. We recommend leaving the bar in situ for three years.

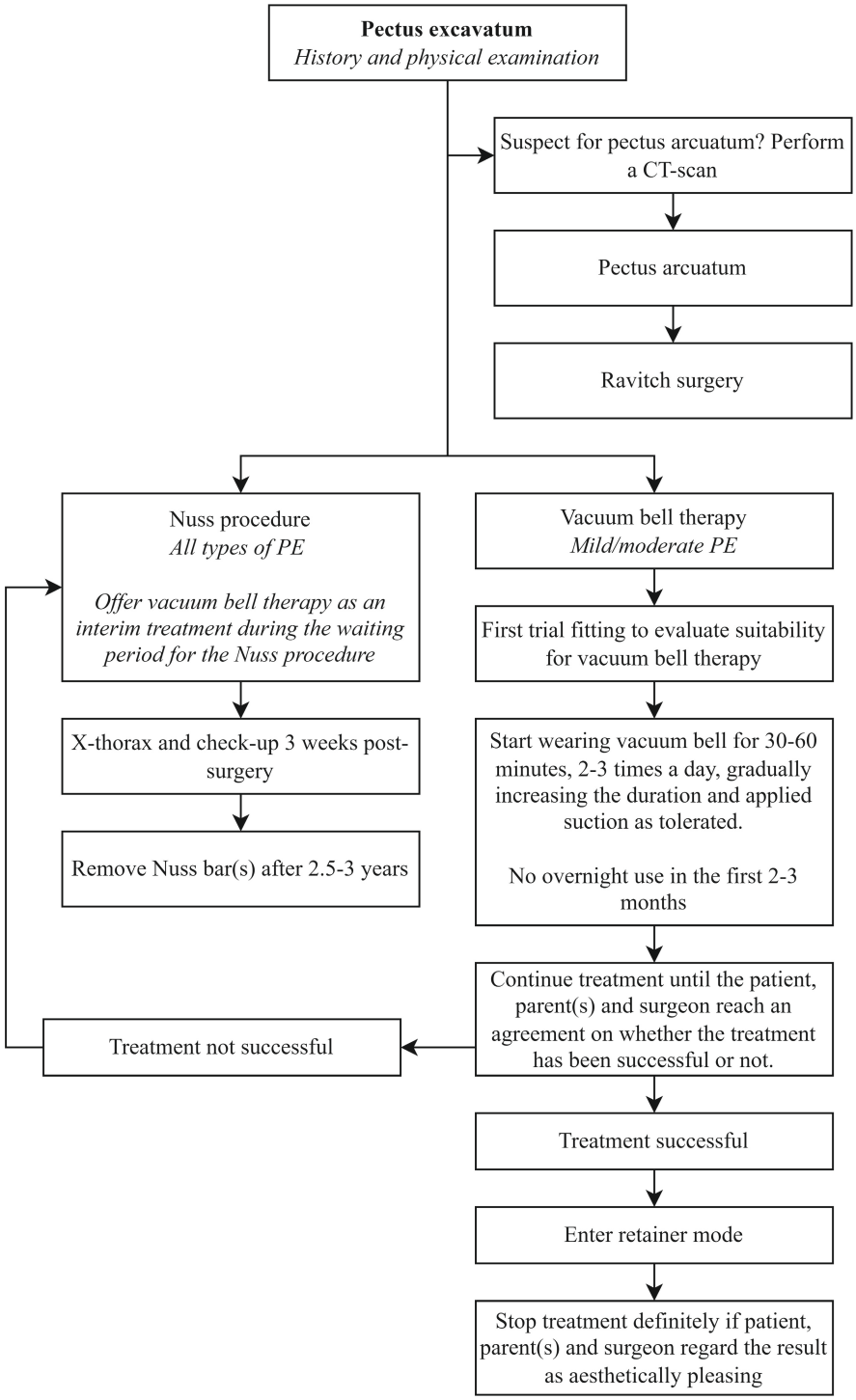


Figure 1 Treatment protocol for pectus excavatum (PE=Pectus Excavatum)

Outcomes

Primary outcomes were incidence and management of Nuss bar infections. Secondary, the early- and late onset infections were compared based on annual incidence, C-reactive protein (CRP), leukocyte count and type of bacteria. Also, the superficial and deep infections were compared based on the need for surgical intervention, hospital admission, wound drainage, type of cultured bacteria, timing of infection, annual incidence, and type and duration of antibiotic treatment. Other secondary outcomes were infection symptoms, used diagnostics, type of cultured bacteria, analysis of prognostic factors for Nuss bar infections (sex, age, type of anesthesia, number of Nuss bars, Nuss bar dislocation, place of stabilizer plates, suture use), Nuss bar removal time after infection and incidence of allergies and terra firma-forme dermatosis.

Statistical analysis

Descriptive measurements were utilized for baseline characteristics. A P-value <0.05 was considered significant. Data were analyzed using IBM SPSS Statistics 28.0. Distribution of continuous variables was assessed using the Shapiro-Wilk test. Normally distributed variables are reported as mean ± standard deviation (SD) and non-normally distributed variables are reported as median with interquartile range (IQR).

RESULTS

All (N=695) patients who underwent the Nuss procedure were included. Median age at surgery was 15.0 (14.0-17.0) years, 82.4% of patients were male (573/695). Most patients received one bar (89.8%, 624/695), 10.1% received two bars (70/695) and 0.1% received three bars (1/695). All patients had a minimal follow-up of six months after Nuss bar placement. Nuss bar removal has occurred in 82.2% of the included patients (571/695), with an median interval of 32.0 (29.0–37.0) months.

Incidence of Nuss bar infections

Of all patients, 4.0% (28/695) developed a Nuss bar infection. Figure 2 shows infections per year and cultured bacteria. There was no relationship between the year of surgery and the incidence of infections (P=.29).

Early versus late-onset infections

The mean time between Nuss bar placement and infection was 27.6±38.7 weeks (6.0±9.0 months). A significant association was found between the year of surgery and infection timing, with early-onset infections predominating in earlier years and a gradual shift toward late-onset infections in recent years (P=.03). Compared to late-onset infections, early-onset infections had higher CRP (mg/L) and leukocyte levels

(10^9L) (CRP 408.0, 240.0, 223.0, 61.8, leukocytes 25.8, 23.0, 15.9, 11.9) (CRP 11.1 (6.2-40.2), leukocytes 9.3 (6.7-10.4)).

Cutibacterium acnes (CA), formerly Propionibacterium acnes, was isolated more frequently in late-onset infections (one versus four), although this difference was not significant (P=.62). Although more early-onset infections occurred in patients with sutures, this difference was not significant (P=.24).

Deep versus superficial infections

Twenty patients (71.4%) developed deep infections. None of the superficial infections required surgery, whereas 55.0% (11/22) of deep infections did (P=.007). No significant differences were seen between deep and superficial infections in hospital admissions (P=.07), wound drainages (P=.14), or bacteria cultured (P=1.0). No correlation was found between deep or superficial infections and the year they occurred (P=.75), early and late-onset infections (P=.62) or antibiotic treatment duration (P=.57).

Symptoms and diagnostics

Table 1 shows an overview of symptoms and diagnostic tests. Most patients presented with erythema (67.9%, 19/28) and pain (71.4%, 20/28), while fever (32.1%, 9/28) and exudate (39.3%, 11/28) were less common. Of patients, 21.4% (6/28) had one symptom, 46.4% (13/28) had two symptoms, and 32.1% (9/28) had three symptoms. Thoracic X-rays and Nuss bar ultrasounds were made in respectively 85.7% (24/28) and 53.4% (15/28) of patients. Allergy tests were conducted in 14.3% (4/28), revealing a cobalt allergy in one patient.

Shift in bacterial species

Figure 2 shows a shift in bacteria over time. CA has replaced Staphylococcus aureus (SA) as the most commonly cultured bacterium since 2018.

Treatment

Table 2 provided an overview of treatment for Nuss bar infections. Of all patients, 53.6% (15/28) was hospitalized one (14/15) or two (1/15) times and 39.3% (11/28) underwent one (8/11) or two (3/11) surgeries. Patients received antibiotics for a mean duration of 51.3±75.0 days. In 14.3% (4/28) the Nuss bar was removed early.

Table 1 Overview of symptoms and (results of) diagnostic tests

	Year	Time after Nuss placement		Early onset	After surgery for dislocation	Symptoms			L	CRP	E	Ultrasound Nuss bar	Deep infection	Thoracic X-ray	Allergy test [a]
						Erythema	Pain	Fever	Exudate							
		Weeks	*Months*												
1	2000	11	2				X						X	X	
2	2001	14	3						X				X	X	
3	2003	3	0	X					X				X	X	
4	2004	1	0	X		X	X		X				X	X	
5	2005	6	1			X	X		X				X	X	
6	2005	11	2		X	X	X	X					X	X	
7	2007	2	0	X		X	X	X		25.8	408.0		X	X	X	
8	2007	11	2		X	X		X	X						X	
9	2008	0	0	X			X	X						X	X	
10	2008	1	0	X			X	X					X	X	X	
11	2009	1	0	X			X	X					X		X	
12	2010	10	2			X				14.0	64.0					
13	2013	130	30			X			X	6.8	6.0	0.13	X	X	X	0
14	2015	2	0	X		X	X		X				X			
15	2016	89	20		X	X	X			5.8	9.5	0.07	X	X		
16	2017	3	0	X		X	X	X		23.0	240.0	0.00	X	X	X	
17	2017	3	0	X	X		X	X		15.9	223.5	0.07		X	X	
18	2018	107	24			X				6.6	8.0		X	X	X	
19	2019	105	24			X	X			10.3	3.0		X		X	0
20	2019	6	1		X		X	X		9.7	47.0		X	X	X	

Table 1 Overview of symptoms and (results of) diagnostic tests *Continued.*

	Year	Time after Nuss placement		Early onset	After surgery for dislocation	Symptoms				L	CRP	E	Ultrasound Nuss bar	Deep infection	Thoracic X-ray	Allergy test [a]
						Erythema	Pain	Fever	Exudate							
		Weeks	*Months*													
21	2019	71	16		X	X	X			8.0	15.0	0.12	X		X	
22	2019	66	15			X	X		X	9.2	0.8		X	X	X	
23	2020	3	0	X		X			X	11.9	61.8	0.57			X	
* 24	2021	24	5		X	X	X			12.9	64.9	0.16	X	X	X	1
25	2021	5	1			X	X			5.5	6.7	0.32			X	0
26	2022	54	12			X	X			10.4	19.6	0.36	X	X	X	
27	2023	28	6						X					X	X	
28	2024	5	1			X	X		X	9.3	12.7		X	X		
N (%)				10 (35.7)	7 (25.0)	19 (67.9)	20 (71.4)	9 (32.1)	11 (39.3)				15 (53.4)	20 (81.4)	15 (85.7)	4 (14.3)
Mean (SD)		27.6 (38.7)	6.0 (9.0)													

CRP=C-Reactive Protein (mg/L), E=eosinophils (10^9/L), L=leukocytes (10^9/L)
[a] 0=negative, 1=positive, * cobalt allergy

Table 2 Overview of treatment

	Year	N of hospital admissions	LOS	N of surgery	Drainage	Tissue culture	Bacterium	Total duration of antibiotics	Intervals of antibiotics (administered sequentially, unless in the same column)				Bar removed early (months)
			Days						*Antibiotic (days of treatment)*				
1	2000	1	1	1		X	-	10	Au (10)				
2	2001	1	2	1		X	-	10	Au (10) Fl (10)				
3	2003				X	X	-	5	Am (5)				
4	2004	1	10	1				10	Au (2)	Fl (8)			1 (replaced)
5	2005	1	4	2		X	SA	>365	Fl (>365)				
6	2005	1	15	1	X	X	SA	26	Au (5)	Fl (7) Ri (7)	Cl (14) Ri (14)		
7	2007	1	23		X	X	SA	35	Au (2) Ge (2)	Fl (21)	Cl (12)		
8	2007	1	21			X	SA	42	Fl (21)	Fl (21)			
9	2008				X			42	Au (13)	Fl (9)	Fl (20)		
10	2008	1	14	1	X	X	SA	49	Fl (7)	Fl (14)	Fl (28)		
11	2009	1	14					14	Au (3)	Va (11) Cefta (11)			
12	2010							14	Au (14)				
13	2013					X	-	14	Au (14)				
14	2015							10	Fl (10)				
15	2016	2	7	2		X	MM	54	Fl (21)	Ci (14)	Co (5)	Co (14)	
16	2017	1	10	1	X	X	SA	42	Fl (14)	Cl (28)			

Table 2 Overview of treatment *Continued*.

	Year	N of hospital admissions	LOS	N of surgery	Drainage	Tissue culture	Bacterium	Total duration of antibiotics	Intervals of antibiotics (administered sequentially, unless in the same column)					Bar removed early (months)
			Days						*Antibiotic (days of treatment)*					
17	2017	1	14	1		X	SA	42	Fl (14)	Cl (28) Ri (28)				
18	2018					X	CA	7	Au (7)					
19	2019							7	Au (7)					24
20	2019	1	5					49	Fl (5)	Fl (42)				12-22[a]
21	2019					X	-	21	Cl (21)					
22	2019	1	6	2		X	CA	220	Au (6)	Au (8)	Cl (10)	Cl (196)		
23	2020					X	CA	56	Fl (56) Cl (56)					
* 24	2021	1	2	1		X	CA	114	Cl (28)	Cl (53)	Au (14)	Cl (5)	Ph (14)	24
25	2021					X	SA	73	Au (7)	Cl (36)	Cl (30)			
26	2022							14	Fl (14)					
27	2023							49	Am (49)					
28	2024					X	CA	42	Fl (7)	Am (28)				

Am=Amoxicillin, Au= Augmentin, CA=Cutibacterium Acnes, Cefta=Ceftazidime, Ceftr=Ceftriaxone, Ci=Ciprofloxacin, Co=Cotrimoxazole, Fl=Flucloxacillin, Ge=Gentamycin, LOS=Length of Stay, MM=Morganelli Morganii, N=number, Ri=Rifampicin, SA=Staphylococcus Aureus, Va=Vancomycin

Grey shaded=intravenous antibiotics, * cobalt allergy, [a] one bar removed at 12, other at 22 months

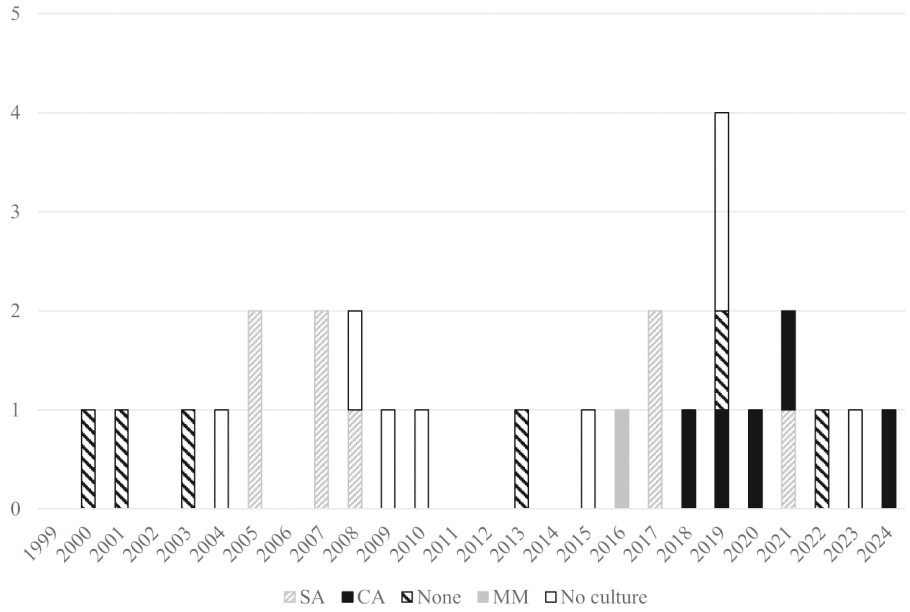


Figure 2 Number of Nuss bar infections per year and cultured bacterial species (CA= Cutibacterium Acnes, MM=Morganelli Morganii, SA=Staphylococcus Aureus)

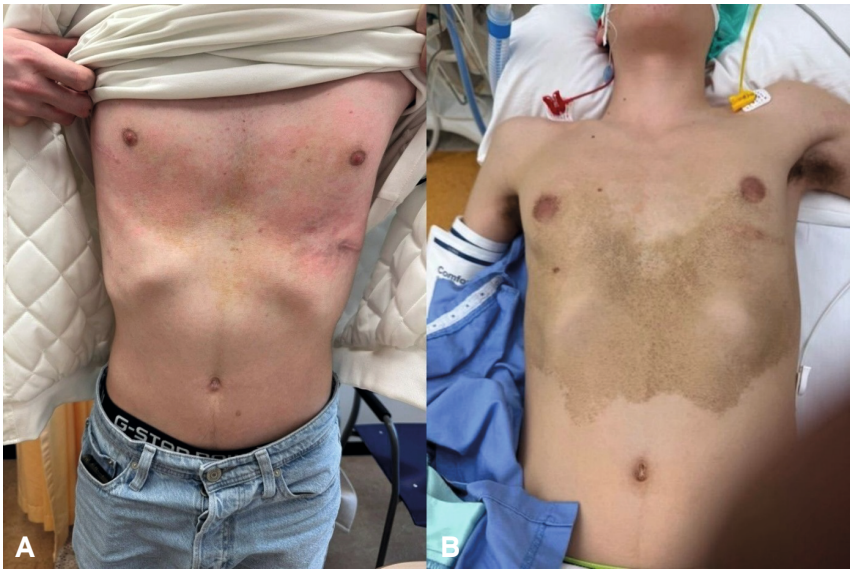


Figure 3 Terra firma-forme dermatosis. Early erythematous lesions (A). Late, brownish, dirt-like patches (B).

Prognostic factors and impact of infection on Nuss bar removal time

Bar dislocation (5.6%, 39/695) and stabilizer plate loosening (1.0%, 7/695), caused reoperations in all affected patients and were more prevalent in Nuss bar infections (P=.001). Reoperations for bar dislocation, plate loosening, and pain have declined over time (P=.02), with an annual rate of 0–3.1% in the past five years.

Stabilizer plates were placed on the left in 60.7% of patients (17/28), on the right in 25.0%, and on both sides in 10.7% (3/28). Infection site and stabilizer plate position were significantly related (P=.007).

Operative time for Nuss bar removal was longer in patients with infections (52.0 (32.0–68.0)), compared to those without (35.0 (26.0–49.0), P=.03). Infection rate was not related to age (P=.06), sex (P=.14), number of bars (P=.15), or type of analgesia (P=.71).

Skin reactions

Aside from bar infections, three patients were suspected allergic. Two experienced allergy-like symptoms, although no allergy was found. One patient's allergy was misdiagnosed as infection. Multiple antibiotics failed to treat his erythema and pain. A cobalt allergy was diagnosed, however prednisolone provided only mild relief. After two years, the patient chose bar removal over methotrexate treatment.

Terra firma-forme dermatosis, presenting as erythematous lesions that subsequently develop into brownish, dirt-like patches resistant to soap and water (Figure 3), occurred in 0.4% of patients (3/695) and was effectively treated with alcohol wipes.

DISCUSSION

Postoperative infections were found in 4.0% of the patients, with a shift from early- to late-onset infections over time. Most common infection symptoms were erythema and pain. CA became increasingly prevalent in the past decade. Infection occurred more often on the stabilizer plate side, and after bar dislocation and bar loosening. Bar removal operation times were longer with bar infections. Allergies can mimic infections. Terra firma-forme dermatosis is effectively treated with alcohol wipes.

Incidence of bar infections

Our infection rate of 4.0%, aligns with current literature (1.5%-9.4%).[3-15] No consensus exists on acceptable infection rates in Nuss bar surgery. Even if the 1-5% threshold for clean wounds excluding prosthetic material is considered, our rate is still within this range, which is acceptable considering prosthetic operations have a higher threshold.

Infection management

Studies describe a wide range of antibiotics, mostly based on the surgeon's preference,[11, 12] with SA being the most commonly cultured bacterium. The latter study suggested oral clindamycin and cotrimoxazole for one to two months as long-term suppressive treatment. Rifampicin was also described in literature.[4] Since SA is common, we recommend oral flucloxacillin plus biofilm-active rifampicin as first-line treatment for acute infections for six weeks. If a pathogen other than SA is found, cultures should be taken and antibiotics changed. Until bar removal, treat low-grade infections with oral amoxicillin or clindamycin. Shared decision-making and bar removal should be considered throughout this phase of treatment due to the risks of long-term antibiotic use. Use intravenous treatment only for clinically unstable individuals.

We recommend the following infection protocol (Figure 4) based on our findings and experience.

Preoperative measures

We recommend cefazolin preoperatively and for 24 hours postoperatively.[4, 22] To minimize infection risk, we recommend following the international guidelines on prevention of surgical site infections.[19, 20] The international guidelines for surgical site infections do not recommend adhesive drapes or double gloves,[20] and recent evidence on the effects of adhesive drapes, including antimicrobial ones, on prosthetic surgery infection rates is inconclusive.[23]

Symptoms, imaging and culturing

Erythema, pain, and exudate indicate infection, but fever is rare. A thoracic X-ray and if inconclusive, a CT scan should be performed in all suspected cases to rule out bar dislocation, pneumonia, and pleural effusion. If these causes are excluded, ultrasound should be used to detect fistulas or fluid collections around the bar. When fluid is present, drainage and culture are essential for guiding antibiotics. Fistulas require surgical treatment.

In all suspected infections, cultures should be obtained before administering antibiotics, preferably fluid aspiration or intraoperative biopsies (at least five).

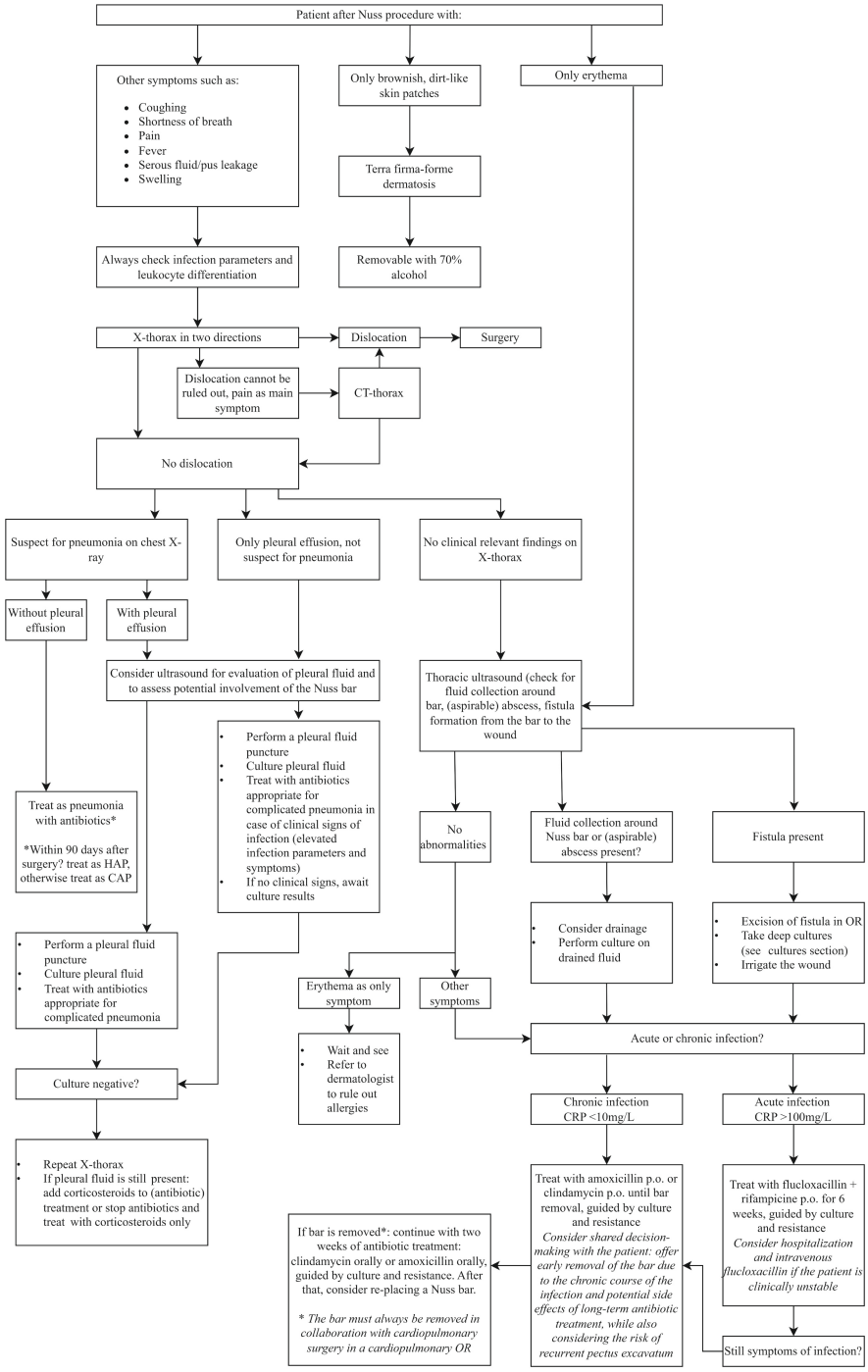


Figure 4 Protocol for managing postoperative Nuss bar infections (CAP = Community Acquired Pneumonia, CRP = C-Reactive Protein, HAP = Hospital Acquired Pneumonia)

Shift in time of infections and type of bacteria

SA was the most commonly cultured pathogen in early-onset Nuss bar infections. Late-onset CA infections, a slow-growing bacterium linked to indolent implant infections, have increased over the past decade. This may be due to improved surgical procedures lowering acute infections and increasing chronic, low-grade infections. Frequent and targeted testing may also increase CA detection. Since CA is a common cutaneous bacterium, it is commonly neglected unless implanted material is present. Surface swabs may detect CA even when it is not clinically relevant, leading to needless antibiotic therapy and delayed allergy diagnosis. Future investigations should focus on culturing explanted bars to identify if CA is a pathogen or a contaminant in chronic infections.

Prognostic factors

Bar dislocation and stabilizer plate loosening were correlated to higher infection rates with 5.6% and 1.0% prevalence, respectively. These findings support previous publications linking mechanical instability to postoperative infection risk.[24] Infections were more frequent on the side with a stabilizer, confirming the idea that enhanced tissue manipulation or injury in these locations may enable bacterial colonization and increase infection risk. Such manipulation may potentially change the local tissue milieu, making latent or low-virulence bacteria like CA clinically relevant.

According to the literature, only the bridge technique has created a zero percent dislocation rate.[25,26] We mostly employ a single bar, making bridging impractical. Despite this, we have lowered our dislocation rate to 0–3.1% during the past five years. This improvement may be attributed to the expanding experience of our surgical team,[27] and the use of screw-fixed stabilizers and their medial placement, which reduces leverage and limits bar movement.

Influence of Nuss bar infection on bar removal

Infections complicate the bar removal procedure, resulting in prolonged operative times. Bar removal and reimplantation after infection require multidisciplinary collaboration, particularly involving cardiac surgeons. The risks of infection and recurrent pectus excavatum should be considered before reimplantation. A second surgery increases the risks due to scar tissue, which increases the risk of pericardial injury. Culture-based two-week targeted antibiotic therapy with oral amoxicillin or clindamycin is recommended prior to bar replacement.

Skin reactions

If prolonged erythema without systemic infection markers, diagnostic imaging abnormalities, or unexplained postoperative inflammation occurs, allergies to implant materials (e.g., cobalt) should be investigated.

A recent article describes delayed-onset postoperative grayish-brown hyperpigmentation,[28] which we believe is terra firma-forme dermatosis. A review noted it occurs more often near surgical sites, possibly triggered by surgery or cryoablation.[29] Though the cause is unclear, the pigmentation resists soap and water but is removed with 70% isopropyl alcohol.

Future directions and limitations

The retrospective nature of our study introduces inherent biases, and may lead to missing data due to non-digitized medical records. The findings of this study can be validated by using standardized follow-up protocols in order to improve techniques.

Additionally, the impact of sutures on infection development requires more research considering the use of hammocks. When sutures were utilized, early infections were more likely, suggesting they fostered bacterial colonization. Fixation methods should be optimized to reduce infection risk as surgical techniques advance.

Conclusion

Postoperative infections had an incidence of 4.0%, with a shift from early- to late-onset infections over time. Bar dislocation and stabilizer plate loosening are correlated with infection, emphasizing the need to optimize surgical procedures. Our infection protocol emphasizes a structured diagnostic approach, tailored antibiotic therapy, and multidisciplinary management. In cases with mild symptoms and negative cultures, or when CA is detected, alternative diagnoses such as implant allergy should be considered. Terra firma-forme dermatosis is easily treated with alcohol wipes.

REFERENCES

1. Nuss D, Kelly RE, Croitoru DP, Katz ME. A 10-year review of a minimally invasive technique for the correction of pectus excavatum. J Pediatr Surg. 1998;33(4):545-52.
2. Aly MR, Farina JM, Botros MM, Jaroszewski DE. Minimally invasive repair of pectus excavatum in adults: a review article of presentation, workup, and surgical treatment. J Thorac Dis. 2023;15(9):5150-73.
3. Obermeyer RJ, Cohen NS, Gaffar S, et al. Multivariate analysis of risk factors for Nuss bar infections: A single center study. J Pediatr Surg. 2018;53(6):1226-9.
4. Obermeyer RJ, Godbout E, Goretsky MJ, et al. Risk factors and management of Nuss bar infections in 1717 patients over 25years. J Pediatr Surg. 2016;51(1):154-8.
5. Coughlin AC, Ahsanuddin S, Inglesby D, et al. "When to Nuss? patient age as a risk factor for complications of minimally invasive repair of pectus excavatum: a systematic review and meta-analysis". Pediatr Surg Int. 2022;38(3):365-75.
6. Park HJ, Lee SY, Lee CS. Complications associated with the Nuss procedure: analysis of risk factors and suggested measures for prevention of complications. J Pediatr Surg. 2004;39(3):391-5.
7. Hebra A, Swoveland B, Egbert M, et al. Outcome analysis of minimally invasive repair of pectus excavatum: review of 251 cases. J Pediatr Surg. 2000;35(2):252-7.
8. Nuss D, Croitoru DP, Kelly RE Jr., et al. Review and discussion of the complications of minimally invasive pectus excavatum repair. Eur J Pediatr Surg. 2002;12(4):230-4.
9. Croitoru DP, Kelly RE Jr., Goretsky MJ, et al. Experience and modification update for the minimally invasive Nuss technique for pectus excavatum repair in 303 patients. J Pediatr Surg. 2002;37(3):437-45.
10. Watanabe A, Watanabe T, Obama T, et al. The use of a lateral stabilizer increases the incidence of wound trouble following the Nuss procedure. Ann Thorac Surg. 2004;77(1):296-300.
11. Shin S, Goretsky MJ, Kelly RE, Jr., Gustin T, Nuss D. Infectious complications after the Nuss repair in a series of 863 patients. J Pediatr Surg. 2007;42(1):87-92.
12. Calkins CM, Shew SB, Sharp RJ, et al. Management of postoperative infections after the minimally invasive pectus excavatum repair. J Pediatr Surg. 2005;40(6):1004-7.
13. Van Renterghem KM, von Bismarck S, Bax NM, Fleer A, Höllwarth ME. Should an infected Nuss bar be removed? J Pediatr Surg. 2005;40(4):670-3.
14. Fallon SC, Slater BJ, Nuchtern JG, et al. Complications related to the Nuss procedure: minimizing risk with operative technique. J Pediatr Surg. 2013;48(5):1044-8.
15. Torre M, Guerriero V, Wong MCY, et al. Complications and trends in minimally invasive repair of pectus excavatum: A large volume, single institution experience. J Pediatr Surg. 2021;56(10):1846-51.
16. Ortega G, Rhee DS, Papandria DJ, et al. An evaluation of surgical site infections by wound classification system using the ACS-NSQIP. J Surg Res. 2012;174(1):33-8.
17. Obermeyer RJ, Cohen NS, Kelly RE Jr., et al. Nonoperative management of pectus excavatum with vacuum bell therapy: A single center study. J Pediatr Surg. 2018;53(6):1221-5.
18. Van Braak H, de Beer SA, de Jong JR, et al. Intercostal Nerve Cryoablation or Epidural Analgesia for Multimodal Pain Management after the Nuss Procedure: A Cohort Study. Eur J Pediatr Surg. 2024;34(6):488-92.
19. Koek MBG, Hopmans TEM, Soetens LC, et al. Adhering to a national surgical care bundle reduces the risk of surgical site infections. PLoS One. 2017;12(9):e0184200.

20. Allegranzi B, Bischoff P, de Jonge S, et al. New WHO recommendations on preoperative measures for surgical site infection prevention: an evidence-based global perspective. Lancet Infect Dis. 2016;16(12):e276-e87.
21. Pilegaard HK. Short Nuss bar procedure. Ann Cardiothorac Surg. 2016;5(5):513-8.
22. M.P. Bauer EMW, M.E.E. van Kasteren, J.M. Prins, M.C. Vos. SWAB Richtlijn Peri-operatieve profylaxe. Leiden: Stichting Werkgroep Antibioticabeleid (SWAB); 2019:19-20.
23. Mundi R, Nucci N, Ekhtiari S, et al. Do Adhesive Drapes Have an Effect on Infection Rates in Orthopaedic Surgery? A Systematic Review and Meta-Analysis. Clin Orthop Relat Res. 2022;480(3):551-9.
24. Media AS, Christensen TD, Katballe N, et al. Complication rates rise with age and Haller index in minimally invasive correction of pectus excavatum: A high-volume, single-center retrospective cohort study. J Thorac Cardiovasc Surg. 2024;168(3):699-711.
25. Park HJ, Kim KS, Moon YK, Lee S. The bridge technique for pectus bar fixation: a method to make the bar un-rotatable. J Pediatr Surg. 2015;50(8):1320-2.
26. Kim H, Rim G, Park HJ. Technical Advances in Pectus Bar Stabilization in Chest Wall Deformity Surgery: 10-Year Trends and an Appraisal with 1,500 Patients. J Chest Surg. 2023;56(4):229-37.
27. Park HJ, Lee S, Lee C. Complications Associated with the Nuss Procedure: Analysis of Risk Factors and Suggested Measures for Prevention of Complications. J Pediatr Surg. 2004;39:391-5.
28. Pitt JB, Carter M, Zeineddin S, et al. Chest Wall Dermatitis Patterns Following Thoracoscopic Intercostal Nerve Cryoablation for Surgical Correction of Pectus Excavatum. J Pediatr Surg. 2024;59(9):1687-93.
29. Sechi A, Patrizi A, Savoia F, et al. Terra firma-forme dermatosis: a systematic review. Int J Dermatol. 2021;60(8):933-43.

Part III

Cross-cutting perspectives

Chapter 10

Time heals: the impact of waiting times on pediatric patients' decisions to decline pectus surgery

Hendrik van Braak, Sjoerd A. de Beer, Matthijs W.N. Oomen, Sander Zwaveling, Joep P.M. Derikx, L.W. Ernest van Heurn, Justin R. de Jong

Journal of Pediatric Surgery. 2025 Nov 60:162504.

ABSTRACT

Background

This study investigates the impact of waiting times, due to Covid-19, on patients' decisions to undergo surgery for pectus deformities.

Methods

We conducted a cross-sectional study of patients (>18 years) on the pectus surgery waitlist at the Amsterdam Pectus Centre. Patients were contacted in January 2025. Primary outcome was the proportion of patients still wanting surgery. Secondary outcomes included symptom progression, reasons and predictive factors for withdrawal (Ravitch surgery, age at waitlist entry, male sex, physical and psychosocial symptoms at waitlist entry). Predictive factors were analyzed using multivariable logistic regression.

Results

Of 141 contacted patients, 75.9% (107/141) were included. Age at waitlist entry was 16.0 years (IQR 15.0-17.0). After an average wait of 56.6 months (SD 22.2), only 52.3% (23/44) of pectus excavatum, 30.9% (17/55) of pectus carinatum/arcuatum and 37.5% (3/8) of flaring patients still wanted surgery. Reasons for withdrawal differed: pectus excavatum patients most often cited body acceptance, while pectus carinatum/arcuatum patients most frequently mentioned strength training. Patients who withdrew showed greater reductions in physical (69.6% versus 25.9%, P=.002) and psychosocial symptoms (95.2% versus 53.8%, P=.004) than patients still wanting surgery. Thirteen patients were treated conservatively while awaiting surgery, with a 35.5% success rate. Age (OR=.53, 95% CI=.33-.86, P=.01) and physical symptoms (OR=.37, 95% CI=.15-.94, P=.04) at waitlist entry were predictors of withdrawal from surgery.

Conclusions

After prolonged waiting, 59.8% of all patients withdrew from surgery, primarily due to body acceptance and physical development. Psychological counseling and strength training should be integrated into pectus care, particularly for young, asymptomatic patients.

INTRODUCTION

Pectus deformities, including pectus excavatum (PE) and pectus carinatum (PC), can impact patients' mental and physical well-being. PC is primarily associated with psychological distress, a disturbed body image, and reduced quality of life (QoL).[1] Unlike PE, no direct link between PC and physical impairment has been established.[2] In contrast, PE has been increasingly linked to cardiopulmonary dysfunction, with recent studies demonstrating improved cardiopulmonary outcomes following surgical correction.[3-5]

Most research in pectus deformities has focused on adults with PE, even though presentation differs by age. Adults usually present with symptomatic PE, most commonly with reduced exercise tolerance and shortness of breath,[6] whereas pediatric patients seldom report physical symptoms before puberty. After puberty, shortness of breath, exercise intolerance and diminished endurance become common.[7, 8] Some patients, however, develop symptoms only well into adulthood, and others realize their earlier limitations only after postoperative improvement.[5, 7, 9] These age-related differences have fueled ongoing debate regarding the necessity and timing of pectus surgery in pediatric patients.

Recently, this discussion was reignited when we observed that numerous patients, who had been on our waitlist for surgery for several years, partly due to delays caused by COVID-19, and had reached adulthood in the meantime, no longer wished to undergo surgery. This suggests that some patients might benefit from postponing the decision to undergo surgery for their pectus deformity until adulthood, when cognitive maturity and a more stable self-image may lead to more informed and deliberate choices regarding surgery.

In light of these findings, we aim to further investigate the impact of waiting times on pectus patients' decisions to proceed with surgical treatment, as well as explore predictive factors that influence these decisions.

PATIENTS AND METHODS

Patients and study design

We conducted a cross-sectional cohort study of patients (>18 years) who were on the waitlist for either the Nuss procedure, Ravitch surgery or flaring correction at the Amsterdam Pectus Centre. These patients were contacted in January 2025. A Medical Ethics Review Committee (METC) official waiver of ethical approval was granted by the METC of the Amsterdam University Medical Center. Informed consent was obtained from all patients (or their parents).

Treatment protocol

Our treatment protocol is shown in Figure 1. We advise patients to begin chest exercises and engage in sports while awaiting surgery. Some patients, although primarily wanting surgery, also initiate conservative therapy while awaiting surgery to help maintain chest wall flexibility. In case of isolated flaring, patients are offered surgical resection. For patients with PE combined with flaring, flaring resection is offered at the time of Nuss bar removal, as the placement of the Nuss bar often alters the appearance of the flaring. For patients with PC combined with flaring, flaring can be removed during Ravitch surgery or after completing Dynamic Compression System (DCS) bracing treatment, for similar reasons as with PE. After electing to undergo surgery, patients are placed on the waiting list according to a first-come, first-served principle. All patients are evaluated by a surgeon prior to being added to the list. There is no standard follow-up during the waiting period unless specifically agreed upon by the surgeon and patient.

Outcomes

The primary outcome of the study was the proportion of patients still willing to undergo surgery. Secondary outcomes included reasons for withdrawing from surgery, symptom progression over time and the identification of predictive factors for withdrawal from surgery (Nuss procedure or Ravitch procedure) after a waiting period.

Data extraction

Patients were contacted by phone by HB to assess their continued interest in undergoing surgery. All patients were asked the same questions (Appendix A). All other data were extracted from patients' medical records. Patients were considered lost to follow-up if there was no response after three phone calls – made at different times of the day, each one week apart – and no response to email after another three weeks.

To assess the severity of the deformity, four standardized medical photographs (example in Figure 2) were taken from different angles as part of our protocol. Three pediatric surgeons independently reviewed and classified the images as mild, moderate, or severe. If reviewer ratings differed no more than one category (e.g. mild, mild, moderate), the median rating was used. Otherwise, consensus was reached through discussion.

Reasons for surgery were categorized into three groups: physical symptoms, psychosocial symptoms, and cosmetic reasons. Physical symptoms and psychosocial symptoms were patient reported. Physical symptoms were categorized as reduced exercise tolerance, pain (posture dependent pain, sharp pain), shortness of breath, dysphagia, pressure-like chest pain, palpitations, back pain, fainting episodes. Psychosocial symptoms were defined as dissatisfaction with the appearance of the thorax, accompanied by an impact on daily life (e.g., avoiding swimming or social activities). Cosmetic reasons were defined as dissatisfaction with the appearance of the thorax without associated psychosocial symptoms.

Conservative therapy included DCS-bracing or Vacuum Bell (VB) therapy. Practical considerations for withdrawing from surgery include factors like lack of time due to work, study commitments, or personal responsibilities.

Statistical analysis

Data were analyzed using IBM SPSS Statistics 28.0. Descriptive measurements were utilized to characterize the study population. Normality of continuous variables was assessed using the Shapiro-Wilk test. Normally distributed variables are reported as mean ± standard deviation (SD) and non-normally distributed variables are reported as median with interquartile range (IQR). Of the available retrospective data, potential predictive factors for withdrawal from surgery (Nuss or Ravitch) were selected based on clinical experience. Patients with only flaring were excluded from this analysis as the indication for this type of surgery is typically established after previous PE or PC treatment and involves different decision-making factors.

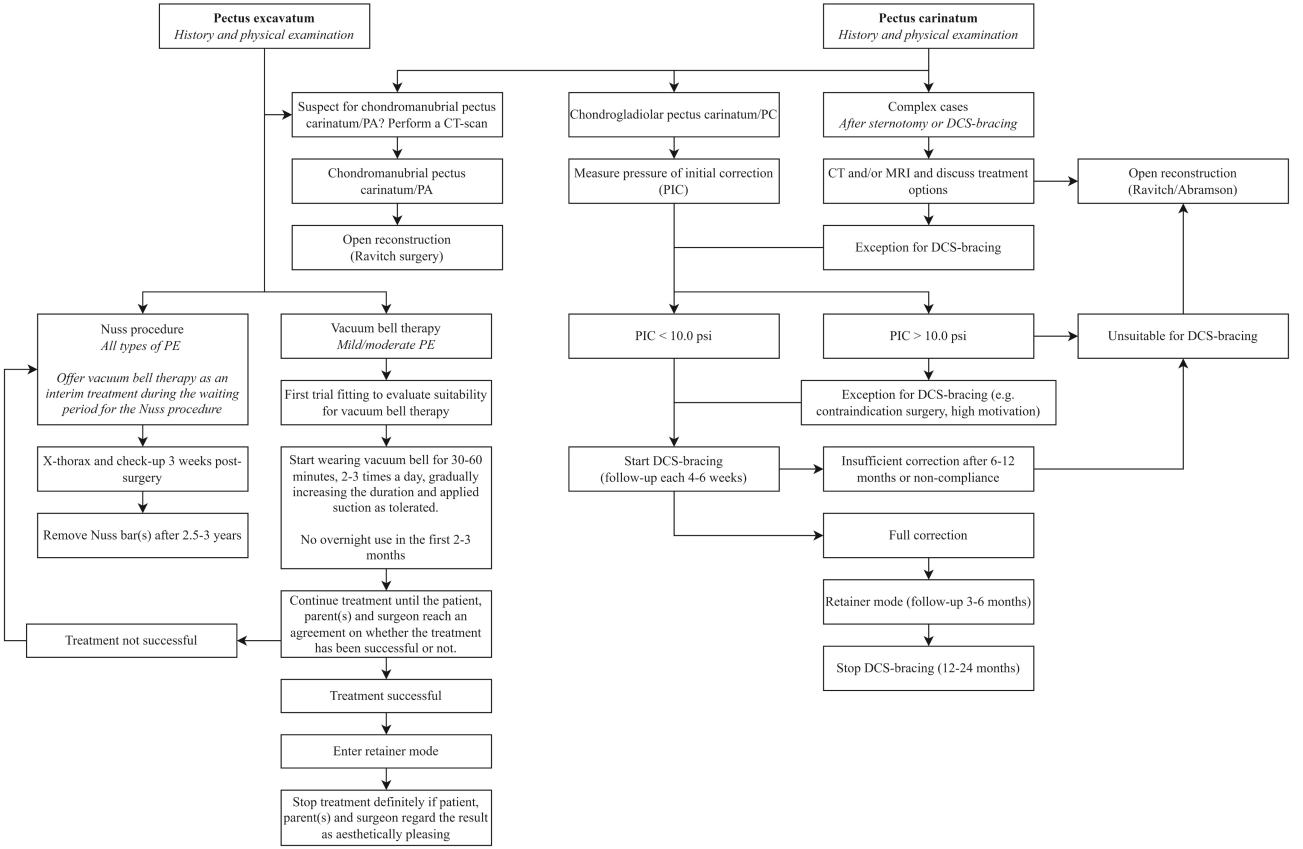


Figure 1 Treatment protocol for pectus excavatum, pectus carinatum and pectus arcuatum (DCS-bracing = Dynamic Compression System Bracing, PA = Pectus Arcuatum, PC = Pectus Carinatum, PE = Pectus Excavatum, PIC = Pressure of Initial Correction)

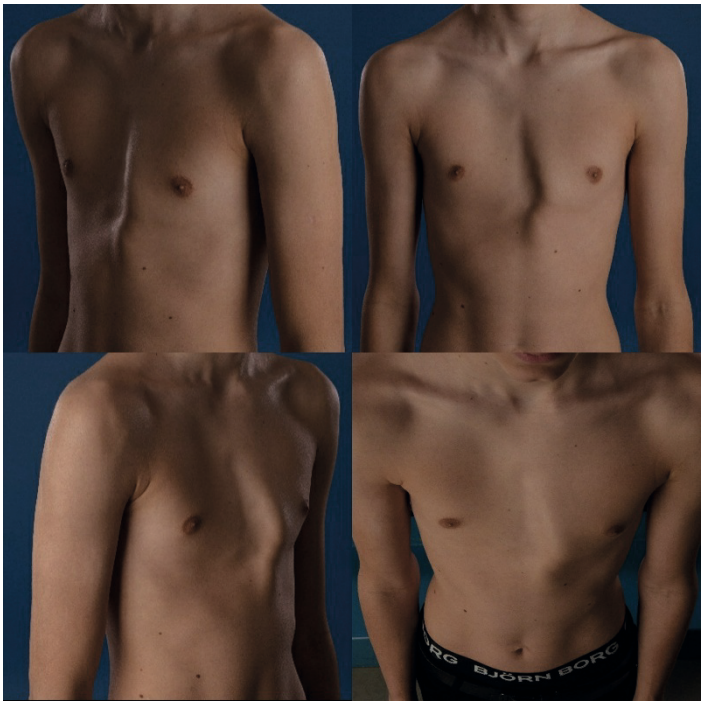


Figure 2 Medical photographs of patient with pectus carinatum

First, univariable logistic regression analysis was performed. Only variables with low collinearity (correlation coefficient between -0.8 and 0.8) were included in the analysis. The input variables included Ravitch surgery, age at waitlist entry, male sex, mild deformity, severe deformity, physical symptoms at waitlist entry and psychosocial symptoms at waitlist entry. Before performing the regression analysis, we examined whether these variables differed significantly between patients listed for Ravitch surgery compared to patients listed for the Nuss procedure, to ensure variable independence. Results were reported in odds ratio (OR) with 95% confidence intervals (95% CI). Nagelkerke's R^2 was calculated to assess the proportion of variation in the outcome explained by the model, with a value >0.15 considered indicative of a meaningful model fit.[10] A receiver-operating characteristic (ROC) curve was created to assess model performance. Area under the curve (AUC) was assessed to quantify the overall discriminative quality of the model. An AUC score of .50–.69 was considered poor, .70–.79 as moderate, and a score of ≥.8 as excellent. Both tests are needed because the ROC curve evaluates the model's discriminative ability, while Nagelkerke's R^2 provides insight into how much of the variability in the outcome is explained by the predictors. A P-value <.05 was considered statistically significant.

RESULTS

As of January 2025, 141 patients (aged >18 years) were on the waitlist for either the Nuss procedure (64/141), Ravitch surgery (67/141) or flaring correction (10/141). Baseline characteristics of the patients can be found in Table 1. Most patients were male (92.2%, 130/141), median age at waitlist entry was 16.0 (15.0-17.0) years and mean waiting time was 56.6±22.2 months (range 11.2-100.0).

Reasons for prolonged waiting time

First, the COVID-19 pandemic significantly increased the surgical backlog. Second, all included patients turned eighteen while waiting. As a pediatric pectus center, we initially could not prioritize adult patients due to staffing shortages, limited operation room availability post-COVID and the elective nature of pectus surgery. Although we offered transfer to other hospitals, most chose to remain on the waiting list until adult pectus surgery resumed.

Severity of deformities, symptoms and motivation for surgery

The severity of the deformity was regarded as mild in 27.4% (32/117), moderate in 63.2% (74/117), and severe in 9.4% (11/117) of patients from for whom medical photographs were available.

Nearly half of patients (45.4%, 64/141) reported physical symptoms at waitlist entry. Among patients with PE, reduced exercise tolerance was most common (29.7%, 19/64), while patients with PC and flaring most frequently reported pain (resp. 25.4%, 17/67 and 30.0%, 3/10). Additionally, psychosocial symptoms were reported by more than a third of patients (35.5%, 50/141).

Despite the prevalence of physical and psychosocial symptoms, a substantial proportion (39.0%) of patients indicated cosmetic concerns as their primary motivation for surgery. The remaining patients reported either physical (33.3%) or psychological (27.6%) symptoms as their main reason for seeking surgical intervention.

In patients with PE, severity of deformity was not significantly associated with the presence of physical complaints overall (P=.16), nor with reduced exercise tolerance specifically (P=.60). Similarly, in patients with PC, no association was observed between severity of deformity and the presence of physical complaints (P=.70).

Percentage of patients still willing to undergo surgery

Of all patients, 9.2% were lost to follow-up (13/141), all other patients were reached by phone. Among them, 6.4% (9/141) had already undergone surgery at another hospital, and 8.5% (12/141) wanted an outpatient consultation with a surgeon before making a decision. Of the remaining patients, 40.2% (43/107) still wanted surgery. This percentage varied across subgroups of patients on the waitlist for the Nuss procedure (52.3%, 23/44), Ravitch surgery (30.9%, 37/55) or flaring resection (37.5%, 3/8) (Figure 3).

Table 1 Baseline characteristics

	Overall (N=141)	**Nuss procedure (n=64/141)**	**Ravitch surgery (n=67/141)**	**Flaring resection (n=10/141)**
Age at waitlist entry (y) [a]	16.0 (15.0-17.0)	16.0 (15.0-17.0)	15.0 (15.0-16.0)	16.0 (14.5-17.0)
Age at evaluation (y) [a]	21.0 (19.0-22.0)	21.0 (19.0-22.8)	20.0 (19.0-22.0)	20.0 (18.8-21.0)
Waiting period (m)	56.6 ± 22.2	58.6 ± 22.8	56.3 ± 21.6	46.1 ± 20.9
Sex [b]				
Male	130 (92.2)	61 (95.3)	62 (92.5)	7 (70.0)
Female	11 (7.8)	3 (4.7)	5 (7.5)	3 (30.0)
Severity of deformity [b] (n=117)				
Mild	32 (22.7)	12 (18.8)	16 (23.9)	4 (50.0)
Moderate	74 (52.5)	37 (57.8)	33 (49.3)	4 (50.0)
Severe	11 (7.8)	4 (6.0)	7 (10.4)	-
Physical symptoms at waitlist entry [b]	64 (45.4)	32 (50.0)	28 (41.8)	4 (40.0)
Type of physical symptoms [b]				
Reduced exercise tolerance	27 (19.1)	19 (29.7)	8 (11.9)	-
Pain	29 (20.6)	9 (14.1)	17 (25.4)	3 (30.0)
Shortness of breath	9 (6.4)	5 (7.8)	4 (6.0)	-
Dysphagia	2 (1.4)	2 (3.1)	-	-
Pressure-like chest pain	6 (4.3)	2 (3.1)	4 (6.0)	-
Palpitations	2 (1.4)	2 (3.1)	-	-
Back pain	2 (1.4)	-	1 (1.5)	1 (10.0)

Table 1 Baseline characteristics *Continued*.

	Overall (N=141)	Nuss procedure (n=64/141)	Ravitch surgery (n=67/141)	Flaring resection (n=10/141)
Fainting episodes	2 (1.4)	1 (1.6)	1 (1.5)	-
Psychosocial symptoms at waitlist entry [b]	50 (35.5)	24 (37.5)	20 (29.9)	6 (60.0)
Main reason for surgery [b]				
Physical symptoms	47 (33.3)	24 (37.5)	21 (31.3)	2 (20.0)
Psychosocial symptoms	39 (27.6)	20 (31.3)	14 (20.9)	5 (50.0)
Cosmetic reasons	55 (39.0)	20 (31.3)	32 (47.8)	3 (30.0)
PIC (PSI, n=34/67)	-	-	7.1 ± 1.8	-
After conservative treatment for PE/ PC [b]	24 (17.0)	6 (9.4)	14 (20.9)	4 (40.0)
Underlying syndrome [b]	4 (2.8)	-	4 (6.0)	-

[a] Continues variables expressed as median (IQR); all remaining continuous variables are expressed as mean ± standard deviation
[b] Data displayed as n (%)
IQR = Interquartile Range, m = months, y = years, n/N = number, PC = Pectus Carinatum, PE = Pectus Excavatum, PIC = Pressure of Initial Correction, PSI = Pounds per Square Inch

Table 2 Reasons for withdrawal from surgery

	Overall (N=64)	Nuss procedure (n=21)	Ravitch surgery (n=38)	Flaring resection (n=5)
Reason for withdrawal [a]				
Acceptance of body	25 (39.1)	11 (52.4)	13 (34.2)	1 (20.0)
Strength training or weight gain	22 (34.4)	1 (4.8)	19 (50.0)	2 (40.0)
Practical considerations	12 (18.8)	6 (28.6)	4 (10.5)	2 (40.0)
Successfully treated with conservative management	5 (7.8)	3 (14.3)	2 (5.3)	-

[a] Data displayed as n (%)
n/N = number

Table 3 Logistic regression for surgery versus no surgery after period on waiting list

Variable	**OR**	**95% CI**	**P-value**
Univariable models [a]			
Ravitch surgery [b]	2.45	1.08-5.57	.03
Age at waitlist entry	.54	.36-.82	.003
Male sex	2.09	.21-20.84	.53
Mild deformity *(n=83)*	2.73	.88-8.41	.08
Severe deformity *(n=83)*	.40	.09-1.80	.23
Physical symptoms at waitlist entry	.31	.13-.72	.006
Psychosocial symptoms at waitlist entry	1.14	.49-2.68	.75
Multivariable model [a]			
Ravitch surgery [b]	1.54	.61-3.90	.36
Age at waitlist entry	.53	.33-.86	.01
Male sex	6.86	.27-174.94	.24
Physical symptoms at waitlist entry	.37	.15-.94	.04
Psychosocial symptoms at waitlist entry	.96	.37-2.5	.94

[a] Analysis in n=99 patients
[b] Reference: Nuss procedure
95% CI = 95% Confidence Interval, n/N = number, OR = Odds Ratio

Reasons for withdrawing from surgery

Patients reported various reasons for withdrawal from surgery (Table 2), most commonly acceptance of their body image (39.1%, 25/107), followed by strength training or weight gain (34.4%, 22/107), practical considerations (18.8%, 12/107), and successful conservative therapy (7.8%, 5/107). Among patients awaiting the Nuss procedure, body acceptance was the primary reason (52.4%, 11/21), followed by practical considerations (28.6%, 6/21). In contrast, patients awaiting Ravitch surgery most frequently cited strength training or weight gain (50.0%, 19/38), with body acceptance ranked second (34.2%, 13/38). Patients awaiting flaring resection reported either strength training or weight gain (40.0%, 2/5) or body acceptance (40.0%, 2/5) as primary reasons for withdrawal from surgery.

Thirteen patients (9.2%) were treated conservatively while awaiting surgery; this was successful in three of eight PE patients (37.5%) and two of five PC patients (40.0%).

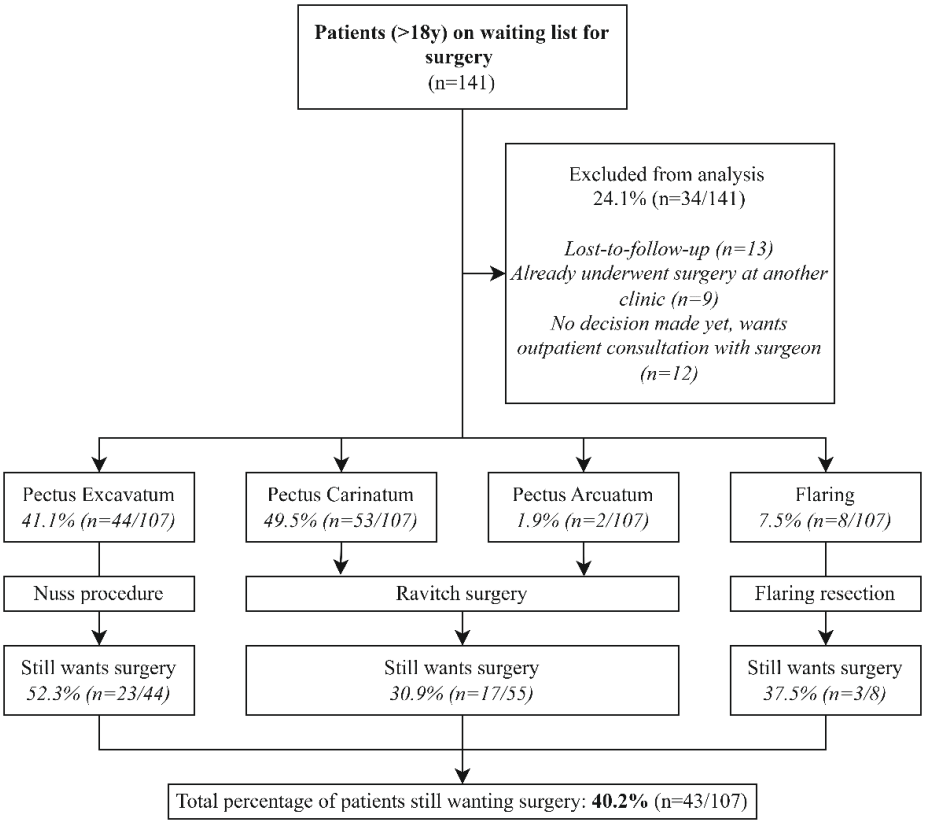


Figure 3 Flowchart of patients included in analysis (n = number, y = year)

Symptom progression over time

The proportion of all patients experiencing physical (from 45.4% to 27.1%) and psychosocial symptoms (from 35.5% to 7.5%) decreased over time (resp. P<.001, P<.001). When comparing patients who withdrew from surgery to those who still wanted surgery, the group withdrawing showed a greater reduction in both physical (69.6% versus 25.9%, P=.002) and psychosocial symptoms (95.2% versus 53.8%, P=.004). Among PE patients, those who withdrew from surgery experienced a larger reduction in physical complaints than those who still wanted surgery (75.0% versus 28.6%, P=.02).

Predictive factors for withdrawal from surgery after a waiting period

Ravitch surgery, age at waitlist entry and physical symptoms at waitlist entry were individually predictive factors of withdrawal from surgery (Table 3). Mild and severe

deformities were included in the univariable model but excluded from the multivariable model due to the proportion of missing values (16/99), which could compromise the integrity and reliability of the model. In the multivariable regression analysis only age at waitlist entry (OR=.53, 95% CI=.33-.86, P=.01) and physical symptoms at waitlist entry (OR=.37, 95% CI=.15-.94, P=.04) remained significant predictors. The final model explained approximately 24% of the variance in outcomes (Nagelkerke's R^2=.24), with an AUC of .74, indicating a clinical meaningful model with a moderate level of discriminative performance (Appendix B).

DISCUSSION

After an average waiting period of nearly five years, only 40.2% of the patients initially listed for any type of pectus surgery still wished to undergo the procedure. Patients withdrew from surgery mainly due to body acceptance or strength training/weight gain, while practical considerations and successful conservative therapy were mentioned less frequently. Reasons varied by deformity type; PE patients primarily cited body acceptance, while strength training or weight gain was the main reason among PC and flaring patients. Conservative treatment offered a moderate success rate during the waiting period. Withdrawal was associated with reductions in both physical and psychosocial symptoms. Younger age and absence of physical symptoms at waitlist entry were predictors of withdrawal from surgery.

Withdrawal from surgery because of body acceptance

Our results show that two-thirds of all patients initially viewed their pectus deformity as a major concern due to cosmetic or psychosocial distress. However, many reconsidered undergoing surgery as they matured and gained greater body acceptance. Moreover, we showed that psychosocial symptoms significantly decreased over time, particularly among patients who withdrew from surgery. Although surgery has been shown to improve self-esteem and body image,[11, 12] our findings suggest that time may also play a crucial role in shifting patients' perspectives regarding body image, self-esteem and the need for surgery.

During adolescence, self-image is still developing, often intensifying body concerns and increasing dissatisfaction with appearance.[13] As cognitive maturity progresses, individuals typically achieve a more stable self-perception, which is reflected by our findings. This process seems to extend into adulthood.[14] In contrast, some authors suggest that body image is largely shaped during a critical period before mid-adolescence, meaning that not all individuals will experience improvement in body image as they mature.[15]

The question is whether surgery should be part of the first treatment options for pediatric pectus patients without clear physical complaints or severe deformity. While pectus deformities may be the cause of lower self-esteem,[16] our findings suggest that surgery is not the only way to address this issue. Rather than focusing solely on chest wall correction, improving self-esteem through targeted psychological support could be an equally effective and less invasive alternative, potentially reducing the need for surgery in mild cases.

However, while this approach may be well suited for patients with PC, it does not necessarily apply to those with PE, since the absence of symptoms does not rule out physical impairment in these patients. Seemingly asymptomatic patients can harbor silent cardiopulmonary limits such as lower stroke volume, reduced VO_2 max and right-ventricular compression, that become evident only on objective testing or after surgical correction.[5] Objective assessment of cardiopulmonary function is essential to determine whether these patients would benefit from surgery.

Withdrawal from surgery because of strength training or weight gain

In addition to psychosocial support, PC patients could also benefit from advice on physical conditioning and muscle development. Similar to our earlier research,[17] this study shows that increasing muscle mass, either through natural growth or training, can effectively conceal PC. Moreover, since most patients are very lean,[18] gaining weight could also be beneficial in masking the deformity.

The necessity of Ravitch surgery should therefore be carefully reconsidered, given that eighty percent of patients initially listed for Ravitch surgery withdrew, due to body acceptance, strength training or weight gain, and concerns about potential surgical risks. Ravitch surgery may be more appropriately reserved for severe cases that are unsuitable for DCS-bracing.

Role of conservative treatment while awaiting surgery

While conservative treatment with the DCS-brace is already standard for most PC patients, the Nuss procedure remains the primary treatment for PE, except for flexible, mild PE. Our previous research demonstrated that VB therapy could be beneficial for patients awaiting surgery,[19] and this study further supports this potential, with 37.5% (3/8) of patients being successfully treated. Although this finding is based on a small subgroup, we believe that VB therapy, combined with psychosocial support, should play a greater role in the treatment of PE if surgery is delayed.

The drawbacks of postponing surgery

Prolonged waiting periods also present practical challenges. Nearly twenty percent of patients who withdrew from surgery cited practical considerations as their reason, indicating that they still desired surgery but could no longer accommodate it in their lives due to competing obligations. This raises concerns about missed treatment opportunities, particularly for those who may have benefitted from surgery but were unable to undergo the procedure due to time constraints rather than a change in perception. For these patients, earlier intervention might have prevented a situation in which surgery is no longer feasible due to life circumstances.

Moreover, with increasing age, progressive ossification leads to greater chest wall rigidity, often requiring additional implants or stabilization techniques to achieve adequate correction in patients with PE. Older age has also been associated with a higher risk of complications.[20] While delaying surgery may help some patients avoid it altogether, it can increase technical complexity and risk for those who eventually undergo the procedure.

Predictive factors for withdrawal from surgery

Age at waitlist entry and the presence of physical symptoms were negative predictive factors for withdrawing from surgery, meaning that the probability of withdrawal decreased as patients were older at waitlist entry and if they had symptoms at that time. In other words, a young, asymptomatic patient on the waitlist has a high chance of changing his/her mind about surgery as they grows older. This finding reinforces the earlier remark that younger patients are still developing their self-image and cognitive maturity, which can shift the perception of their deformity over time. As they grow older, they not only achieve a more stable self-perception but also undergo physical changes, such as the development of muscle mass, which may help conceal the deformity. This natural progression may explain why younger, asymptomatic patients are more likely to withdraw from surgery over time, highlighting the role of psychological adaptation and physical development in decision-making. For the young patient without significant physical symptoms, delaying surgery in favor of psychological and physical support may be a more appropriate initial approach.

The final model demonstrated a moderate ability to distinguish between patients who withdrew from surgery and those who did not. Approximately one-quarter of the variance in withdrawal decisions was explained by the included predictors, making it a meaningful model by current standards. While this leaves room for unmeasured factors, the model proves to be clinically meaningful and is useful in guiding patients through the decision-making process.

Limitations

This study was conducted at a single specialized pectus center, which may limit the generalizability of our findings to other institutions with different treatment protocols or patient populations. However, as a high-volume center with extensive experience in treating pectus deformities, our findings still provide valuable insights into treatment approach.

As a retrospective cohort study, data collection relied on medical records and patient-reported outcomes obtained through telephone interviews, introducing the potential for bias and missing data. We aimed to minimize bias by standardizing the interview process. Due to missing medical photographs, the variables mild and severe deformities could not be included in the multivariable model.

Additionally, surgical indication for PE is typically based on clinical history, visual assessment, and the surgeon's experience. While this long-standing approach is well established in our clinic, it may lack specificity and reproducibility. By contrast, we do use objective measures, such as pressure of initial correction, in patients with PC to support treatment decisions.

Although we observed a marked decline in reported symptoms, particularly among patients who withdrew from surgery, this trend should be interpreted cautiously in patients with PE. Earlier work shows that seemingly asymptomatic PE patients only recognize their cardiopulmonary limitation after experiencing postoperative improvement; thus, absence of self-reported complaints does not guarantee the absence of physiologic impairment.[5] Identifying such patients requires objective assessments, such as cardiopulmonary function testing.

Our findings suggest that psychological support may be an effective alternative to surgery, particularly for younger patients. However, we do not have a clear recommendation on the optimal age on which surgery should be performed. Our study does not define a specific age threshold for when psychological support, combined with conservative treatment, may be sufficient or when surgery becomes the preferred option. We still need to evaluate the implementation of these recommendations in our clinic and plan to report on the outcomes in the future.

We did not evaluate the role of parental expectations and concerns in the decision-making process. Parents play a key role in either reinforcing or alleviating concerns about the pectus deformity in young patients. However, as these patients reach adulthood, they must make the decision independently, which might have influenced

their decision. Although we did not explicitly ask patients about this, we observed that a significant number of adult patients still consulted their parents, suggesting that parental influence may extend beyond childhood.

Conclusions

After an average waiting period of nearly five years, only 40.2% of patients initially listed for surgery still wished to proceed to surgery. Notably, while cosmetic and psychosocial concerns are the main reasons for seeking surgery, improvements in these same areas, through body acceptance or strength training/weight gain, are also the most common reasons for withdrawal. This highlights the importance of integrating psychological counseling into the standard treatment for all pediatric pectus patients, especially the young and asymptomatic patients. This can potentially reduce the need for surgery. Additionally, patients with PC should be encouraged to engage in structured strength training.

For patients with PE, however, caution is warranted: while they may appear asymptomatic, the absence of reported symptoms does not necessarily exclude underlying cardiopulmonary impairment. Furthermore, delaying surgery in PE may increase technical complexity and risk of complications.

REFERENCES

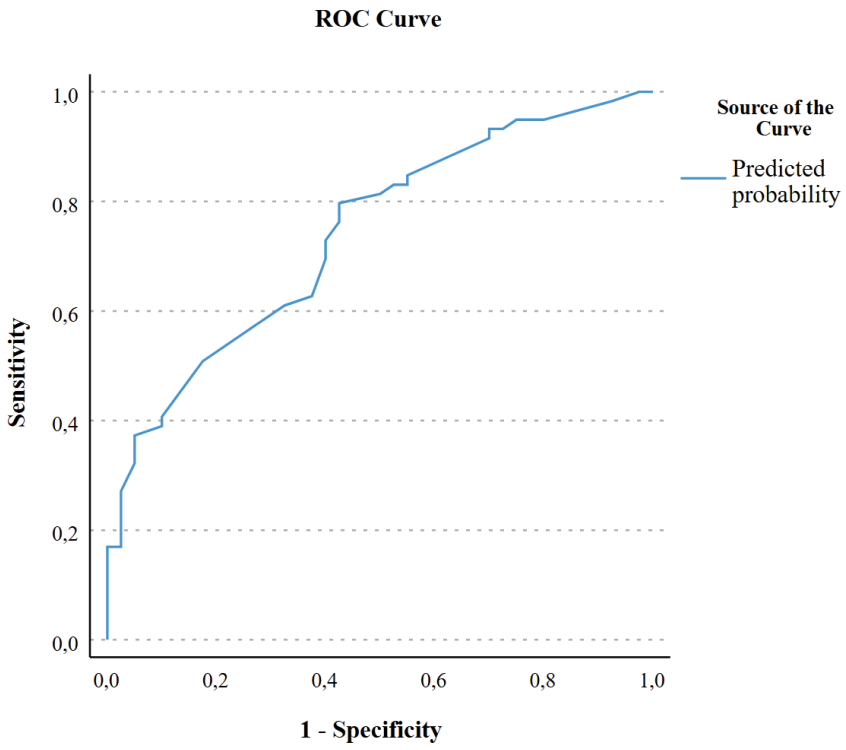
1. Van Braak H, de Beer SA, Twisk JWR, et al. Improving Quality of Life With Dynamic Compression Bracing in Patients With Pectus Carinatum. J Pediatr Surg. 2024;60(1):161975.
2. Orrick BA, Pierce AL, McElroy SF. Changes in self-image after pectus carinatum brace treatment. J Pediatr Surg. 2022;57(8):1579-83.
3. Jaroszewski DE, Farina JM, Gotway MB, et al. Cardiopulmonary Outcomes After the Nuss Procedure in Pectus Excavatum. J Am Heart Assoc. 2022;11(7):e022149.
4. Obermeyer RJ, Cohen NS, Jaroszewski DE. The physiologic impact of pectus excavatum repair. Semin Pediatr Surg. 2018;27(3):127-32.
5. Raggio IM, Toselli L, Valle M, et al. Cardiac decompression and right ventricular function improvement after bar removal in patients with pectus excavatum. Int J Cardiovasc Imaging. 2024;40(11):2383-90.
6. Jaroszewski DE, Aly MR, Shawwaf KA, et al. Phoenix Comprehensive Assessment of Pectus Excavatum Symptoms (PCAPES). J Thorac Dis. 2024;16(12):8565-81.
7. Notrica DM, McMahon LE, Jaroszewski DE. Pectus Disorders: Excavatum, Carinatum and Arcuatum. Adv Pediatr. 2024;71(1):181-94.
8. Janssen N, Coorens NA, Franssen A, et al. Pectus excavatum and carinatum: a narrative review of epidemiology, etiopathogenesis, clinical features, and classification. J Thorac Dis. 2024;16(2):1687-701.
9. Gonzalez GC, Berazaluce AMC, Jenkins TM, et al. Does age matter? Cardiopulmonary profiles of adolescents versus adults with pectus excavatum presenting for surgical evaluation. Pediatr Surg Int. 2022;39(1):52.
10. Gupta A, Stead T, Ganti L. Determining a Meaningful R-squared Value in Clinical Medicine Acad Med. 2024.
11. Zuidema WP, Oosterhuis JWA, Zijp GW, et al. Early Consequences of Pectus Excavatum Surgery on Self-Esteem and General Quality of Life. World J Surg. 2018;42(8):2502-6.
12. Fortmann C, Petersen C. Surgery for Deformities of the Thoracic Wall: No More than Strengthening the Patient's Self-Esteem? Eur J Pediatr Surg. 2018;28(4):355-60.
13. Bucchianeri MM, Arikian AJ, Hannan PJ, Eisenberg ME, Neumark-Sztainer D. Body dissatisfaction from adolescence to young adulthood: findings from a 10-year longitudinal study. Body Image. 2013;10(1):1-7.
14. Gattario KH, Frisén A. From negative to positive body image: Men's and women's journeys from early adolescence to emerging adulthood. Body Image. 2019;28:53-65.
15. Wang SB, Haynos AF, Wall MM, et al. Fifteen-Year Prevalence, Trajectories, and Predictors of Body Dissatisfaction From Adolescence to Middle Adulthood. Clin Psychol Sci. 2019;7(6):1403-15.
16. Steinmann C, Krille S, Mueller A, et al. 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. Eur J Cardiothorac Surg. 2011;40(5):1138-45.
17. Van Braak H, de Beer SA, Zwaveling S, et al. Evaluating Inter- and Intraobserver Agreement on Pectus Carinatum Severity and Treatment Outcomes: A Comparison of Subjective and Objective Assessment Methods. Eur J Pediatr Surg. 2024.
18. Kuru P, Cakiroglu A, Er A, et al. Pectus Excavatum and Pectus Carinatum: Associated Conditions, Family History, and Postoperative Patient Satisfaction. Korean J Thorac Cardiovasc Surg. 2016;49(1):29-34.

19. Van Braak H, de Beer SA, al Ghouch Y, et al. 15 Years of Vacuum Bell Therapy for Pectus Excavatum: Long-term Outcomes and Influencing Factors. J Pediatr Surg. 2025;60(2):161891.
20. Coughlin AC, Ahsanuddin S, Inglesby D, et al. "When to Nuss? patient age as a risk factor for complications of minimally invasive repair of pectus excavatum: a systematic review and meta-analysis". Pediatr Surg Int. 2022;38(3):365-75.

APPENDIX A

Questions
Q1 *What was your initial reason for wanting surgery?*
Q2 *Do you still want to undergo surgery?*
Q3 *If you are no longer interested, what is the main reason for your decision?*
Q4 *Did you experience any physical complaints at the time of your initial assessment?*
Q4.1 *If yes, what complaints did you experience?*
Q4.2 *Do you still experience these physical complaints?*
Q5 *Did you experience any psychological complaints at the time of your initial assessment?*
Q6.1 *If yes, what complaints did you experience?*
Q6.2 *Do you still experience these psychological complaints?*

APPENDIX B


ROC Curve
Sensitivity
1 - Specificity
0,0
0,2
0,4
0,6
0,8
1,0
Source of the Curve
Predicted probability

Part IV

Summary and appendices

Chapter 11

Summary and discussion

SUMMARY

Pectus carinatum (PC) and pectus excavatum (PE) are chest wall deformities that can be treated successfully with both conservative and surgical approaches, each yielding good long-term outcomes. **Part I** focused on PC, comparing Dynamic Compression System (DCS) bracing to Ravitch surgery and demonstrating the advantages of DCS-bracing as the preferred first-line treatment. It also explored the positive impact of bracing on quality of life, highlighted the need for objective outcome measures, and evaluated the potential of 3D scanning for diagnosing PC and monitoring treatment progress. **Part II** addressed PE, assessing the effectiveness of vacuum bell (VB) therapy, comparing pain management strategies following the Nuss procedure, and providing practical guidelines for managing postoperative complications such as pleural effusion, postoperative infections, and allergic reactions. **Part III** examined the impact of surgical wait times on treatment withdrawal among pediatric patients, identifying key predictors of withdrawal and underscoring the importance of psychological factors in treatment decision-making.

PART I: PECTUS CARINATUM

In **Chapter 2**, we compared the outcomes of DCS-bracing and Ravitch surgery in 738 patients with pectus carinatum over a 10-year period. The results showed that DCS-bracing should be the preferred first-line treatment due to a combination its non-invasive nature, good success rate, and lower complication risk compared to surgery. For patients with high initial compression pressures and in those with syndromic conditions such as Marfan and Poland syndrome, however, treatment success of DCS-bracing appeared lower compared to surgery, and poor compliance remained a major challenge in achieving optimal outcomes. Surgical correction, particularly Ravitch surgery, thus remains a valuable alternative for patients with poor bracing prognosis.

In addition to evaluating ability of DCS-bracing to correct PC, in **Chapter 3**, we also report a study that examined whether treatment improved quality of life by assessing the impact of DCS-bracing on quality of life in 225 PC patients aged 10–21 years. The results showed significant improvements in physical complaints, pain, psychological health, self-esteem, and overall quality of life across multiple validated questionnaires, particularly within the first 6–12 months of treatment. No improvement was observed in patients with unsuccessful treatment outcomes. Nearly 90% of all patients would choose bracing again if given the choice, and 95% were satisfied with the treatment.

Despite high satisfaction rates, the absence of standardized objective endpoints continues to complicate DCS-brace treatment. This is addressed in **Chapter 4**, which reports assessment of inter- and intraobserver agreement in the visual evaluation of PC using standardized photographs of 201 patients before and after treatment. Both surgeons and peers showed inadequate consistency in rating esthetic outcomes, severity, and symmetry, with only slight to moderate agreement. Ravitch-treated deformities were perceived as more severe but yielded better esthetic results, while peers rated scars more negatively than surgeons. No associations were found between outcomes and age or treatment duration. These findings underscore the limitations of subjective evaluation and advocate the use of objective measurement tools to assess treatment outcomes.

In response to the need for objective assessment tools, we explored the potential of 3D body imaging as a standardized method to support clinical decision-making in **Chapter 5**. In a cross-sectional study, we compared the External Haller Index (EHI) of 43 patients with PC to that of 50 healthy adolescents. While EHI was significantly lower in patients with PC, the considerable overlap between patients with PC and healthy peers limits its use as a strict diagnostic cut-off. The observed correlation between EHI and Body Mass Index (BMI) suggests that unadjusted EHI values alone are insufficient for reliably monitoring deformity severity or treatment progress, highlighting the need for BMI-adjusted reference values and further validation of 3D imaging parameters.

PART II: PECTUS EXCAVATUM

In **Chapter 6**, we investigated the long-term outcomes and factors influencing the success of vacuum bell (VB) therapy in 259 patients (<18 years) with PE. Among those who completed treatment, success rate was 52.1%. A higher success rate was associated with longer total treatment duration, greater daily use, and overnight application. Deeper deformities, flexible chest walls, and symptomatic presentation were associated with lower success rates. Complications were mostly minor, and recurrence was rare. Notably, 26.7% of patients treated while awaiting a Nuss procedure no longer required surgery. Female patients faced specific challenges, with 39.3% discontinuing treatment due to breast development. Early initiation or watchful waiting may be a more appropriate treatment in this group. These findings underline the need for individualized treatment planning based on patient characteristics to improve outcomes with VB therapy.

The majority of patients with PE are treated with the Nuss procedure. Prior to the introduction of intercostal nerve cryoablation, this minimally invasive surgery was associated with significant postoperative pain, often resulting in extended hospital stays and high opioid consumption. In **Chapter 7** we compared the effectiveness of intercostal nerve cryoablation to continuous epidural analgesia (CEA) in 66 children undergoing the Nuss procedure for PE. The results demonstrated that cryoablation significantly reduced postoperative pain scores, shortened hospital length of stay, and led to markedly lower opioid use both at discharge and one week postoperatively. These findings establish cryoablation as a superior analgesic strategy compared to CEA. Additionally, the data suggest that routine prophylactic use of gabapentin may be unnecessary, given its limited added benefit in patients receiving cryoablation.

Although the Nuss procedure generally yields good results, it is associated with complications, one of which is pleural effusion, a poorly understood and frequently misdiagnosed condition. In **Chapter 8**, we presented three unique, illustrative cases of pleural effusion following the Nuss procedure, showing that in the absence of bar infection or bar displacement, the effusion may represent a reactive inflammatory response that mimics pneumonia, and may respond well to corticosteroid therapy. These cases underscore the importance of thorough diagnostic evaluation and support the use of pleural fluid analysis to guide management. Based on our experience with this complication, we proposed a diagnostic and treatment protocol to help clinicians recognize and manage this complication more effectively.

Another complication of the Nuss procedure is postoperative infection. In **Chapter 9**, we analyzed the incidence, characteristics, and management of Nuss bar infections in 695 patients treated between 1999 and 2024, identifying an overall infection rate of 4.0%. Infections typically presented with erythema, pain, and exudate. Over time, infections shifted from early- to late-onset, with most being deep infections, requiring surgical intervention. Superficial infections were managed with antibiotics, though treatment strategies varied. Bar dislocation and stabilizer plate loosening were significant risk factors for infection. Infections also prolonged bar removal procedures. Based on our findings, we developed a structured diagnostic and treatment protocol, recommending six weeks of oral flucloxacillin and biofilm-active rifampicin for acute infections, and long-term amoxicillin or clindamycin for low-grade cases until bar removal. In patients with persistent erythema but no systemic signs or abnormal imaging, implant allergies should be considered. We also highlighted terra firma-forme dermatosis as a benign postoperative skin condition easily treated with alcohol wipes.

PART III: CROSS-CUTTING PERSPECTIVES

In **Chapter 10**, we investigated the impact of prolonged waiting times, largely due to the COVID-19 pandemic, on patients' decisions to proceed with surgery for pectus deformities. In a cohort of 107 patients, only 40.2% still wished to undergo surgery after an average wait of nearly five years. Withdrawal rates differed by deformity type and were primarily driven by increased body acceptance in patients with PE and by physical development through strength training or weight gain in those with PC or flaring. Patients who withdrew reported greater reductions in both physical and psychosocial symptoms than those who remained on the waitlist. Younger age and absence of physical symptoms at the time of waitlist entry were significant predictors of withdrawal. Conservative treatment was successful in a minority of cases during the waiting period. These findings highlight the importance of early psychological counseling and physical training as part of routine care for young, asymptomatic patients, potentially reducing the need for surgery in selected cases.

DISCUSSION

This thesis explores the current management and future directions in the treatment of patients with pectus deformities. Pectus excavatum (PE) and pectus carinatum (PC) are the most common congenital chest wall deformities and can significantly impact physical function, psychological well-being, and quality of life.[1] Despite advancements in both surgical and conservative treatment modalities, several challenges remain. These include identifying objective criteria for treatment success, managing complications effectively, and improving patient-centered decision-making. This thesis addresses these themes across nine chapters, which can be categorized into three parts. **Part I** evaluates treatment outcomes and quality of life in PC patients, and explores the need and development of 3D body scanning as an objective tool to guide and monitor non-invasive treatment. **Part II** focuses on PE, evaluating (VB) therapy, comparing pain management after the Nuss procedure, and offering practical guidelines for managing complications like pleural effusion, postoperative infections, and allergic reactions. Finally, **Part III** examined the impact of surgical wait times on treatment withdrawal among pediatric patients, identifying key predictors of withdrawal and underscoring the importance of psychological factors in treatment decision-making.

In this chapter we discuss the main findings of our thesis and outline directions for future research.

PECTUS CARINATUM

In **Part I** of this thesis, we compared the outcomes of Ravitch surgery and Dynamic Compression System (DCS) bracing in patients with PC, assessed the impact of DCS-bracing on quality of life, and explored the need for objective treatment endpoints, highlighting the potential of 3D scanning as a tool to facilitate standardized, non-invasive assessment and treatment monitoring.

Before the introduction of the DCS-brace, Ravitch surgery was considered the standard of care for patients with PC. Although DCS-bracing gradually became the preferred first-line treatment due to its non-invasive nature, comparative data on its effectiveness versus surgical correction to date remained limited. In **Chapter 2**, we addressed this gap by directly comparing DCS-bracing to Ravitch surgery in a large cohort. The results showed that while surgery offered slightly better outcomes (92.4% versus 73.8% of patients treated successfully), DCS-bracing should be treatment of first choice because of its non-invasiveness, very few complications and high patient satisfaction. However, there are some exceptions. Bracing should not be initiated before the pubertal growth spurt, as the risk of relapse is higher and long-term retainer use is required, demanding

sustained motivation. Similarly, in patients with a high Pressure of Initial Correction (PIC) (>8.5 PSI), treatment success depends heavily on compliance; if motivation is lacking, outcomes are likely to be poor.[2] Finally, in patients with syndromes such as Marfan or Poland syndrome, or in cases of anatomically complex deformities, Ravitch surgery may be more appropriate than bracing.

Given that treatment outcomes are closely tied to consistency of use, the success rate of DCS-bracing might increase even further if patient compliance could be further improved through more structured and standardized follow-up using, for example, 3D body imaging and digital adherence tracking. Future research should therefore explore interventions aimed at improving compliance, especially in adolescents, who may struggle with long-term treatment commitment.

We do not recommend the Abramson procedure[3] for pediatric PC patients unless specifically requested, as most eligible candidates can also be managed non-operatively with DCS-bracing, thereby avoiding the risks associated with two surgical interventions.

Beyond correction of the physical deformity, we aimed to determine whether treatment with DCS-bracing also resulted in improvements in quality of life for patients with PC, an aspect of pectus deformity care that had not been investigated prior to our study. Therefore, in **Chapter 3** we aimed to investigate the impact of DCS-bracing on quality of life in patients with PC. The results of this study show significant improvements in physical complaints, pain, psychological well-being, self-esteem, and overall quality of life, particularly within the first 6–12 months of treatment.

The results of this study show that patients with PC do profit from DCS-bracing. However, our study had a follow-up of only 24 months. In light of the findings in Chapter 10, it would be valuable to examine whether the distinction between successfully and unsuccessfully treated patients remains relevant in the long term. As some individuals may no longer perceive a need for treatment in adulthood, due to increased body acceptance or physical development, the gap in quality of life between both groups may diminish or even disappear altogether.

Despite the good results of DCS-bracing, determining when treatment can be considered successful remains a challenge. As discussed in **Chapter 4**, current evaluation methods, based on subjective visual assessment, are inconsistent and show poor inter- and intraobserver agreement among both surgeons and peers. To address this, **Chapter 5** explored the potential of 3D body imaging as an radiation free, objective tool to support treatment monitoring and establishing objective outcomes measures. We compared

the External Haller Index (EHI) between PC patients and healthy adolescents. The results show a significant difference in EHI between healthy adolescents and peers, but there was substantial overlap in EHI between the two groups and both cohorts were rather small. While 3D body imaging appears to be a promising non-invasive method for treatment monitoring,[4-7] further research is essential to fully establish its clinical utility. First, a larger cohort of healthy controls is needed to create robust, age- and sex-specific reference values. In addition, development of a BMI-adjusted External Haller Index (similar to existing adjustments in PE care[8]) is critical, as our results demonstrate that BMI significantly influences EHI values. Furthermore, longitudinal studies should assess whether EHI can reliably track deformity progression and treatment response over time. In the future, these measurements could even be integrated into predictive models for treatment planning, optimizing the duration and intensity of bracing therapy. Finally, although EHI represents a useful parameter, it need not be the only or optimal option; alternative indices could be developed that more accurately characterize the deformity and its clinical implications.

PECTUS EXCAVATUM

In **Part II** of this thesis we focused on PE, assessing the effectiveness of VB therapy, comparing intercostal nerve cryoablation to epidural analgesia for postoperative pain management following the Nuss procedure, and providing guidance for the diagnosis and treatment of complications such as pleural effusion, postoperative Nuss bar infections, and allergies.

In **Chapter 6**, we evaluated the long-term outcomes of vacuum bell (VB) therapy as a non-invasive alternative to surgical correction in patients with PE. Among patients who completed treatment, the success rate was 52.1%, which is consistent with outcomes reported in the available literature.[9] Importantly, complications were rare and mostly minor, and recurrence occurred in only 2.3% of patients during long-term follow-up. These findings support VB therapy as a viable treatment option in several patient groups.

First, patients awaiting surgery. In our cohort, 26.7% of patients who initially chose VB therapy while waiting for a Nuss procedure ultimately avoided surgery altogether. Although the numbers are small, this suggests that VB therapy can be a useful interim or even definitive treatment. The second group that might profit from VB therapy are younger patients not yet eligible for surgery. VB therapy may serve as a temporizing measure to preserve thoracic flexibility and prevent progression of the deformity in patients who are too young for surgical correction. However, this requires careful monitoring. The third group that might profit from VB therapy are highly motivated

individuals not willing to undergo surgery. As with DCS-bracing, motivation and compliance are essential. In our study, patients who adhered to longer treatment durations and overnight use had significantly better outcomes. If a patient is highly motivated, a stiff chest wall, contrary to what is generally reported in the literature,[10, 11] should not be considered a contraindication for VB therapy. Notably, female sex should indeed be considered a relative contraindication for VB therapy.

The primary limitation of VB therapy, comparable to DCS-bracing, is patient compliance. In our study, 17.6% of patients were lost to follow-up, and it is likely that treatment adherence was suboptimal in many others. This underscores once again the need of developing improved strategies to support adherence in the conservative management of pectus deformities. As mentioned before, 3D body imaging could play a significant role in this regard, by offering objective and visual feedback on treatment progress, which may help to maintain patient motivation and improve compliance.

In **Chapter 7** we evaluated the effectiveness of intercostal nerve cryoablation compared to epidural analgesia as postoperative pain management strategies following the Nuss procedure. Prior to the introduction of intercostal nerve cryoablation, this minimally invasive surgery was associated with significant postoperative pain, often resulting in extended hospital stays and high opioid consumption. Our results show that intercostal nerve cryoablation significantly reduced postoperative pain scores, shortened hospital length of stay, and led to markedly lower opioid use both at discharge and one week postoperatively. These findings establish intercostal nerve cryoablation as a superior analgesic strategy compared to epidural analgesia. As a result, it has now been adopted as the standard method of analgesia for the Nuss procedure in our center, with epidural analgesia reserved only for selected cases where cryoablation is not feasible (e.g., when single-lung ventilation cannot be achieved).

Yet, the technique continues to evolve. While cryoablation was applied for two minutes per intercostal nerve in this study, newer protocols using a one-minute application are currently being explored.[12, 13] In addition, percutaneous cryoablation, which is already used but not yet widely implemented in clinical practice,[14-16] can potentially further reduce hospital stay and broaden the applicability of the technique, for instance in the treatment of rib fractures or in patients not eligible for thoracoscopic access.

While the Nuss procedure generally yields favorable outcomes and the technique continues to evolve, complications remain an important concern. In **Chapter 8**, we described a case series of patients who developed pleural effusion following the procedure, an underrecognized and frequently misdiagnosed complication that remains

poorly understood. This condition is often mistaken for pneumonia due to overlapping symptoms such as fever, chest pain, dyspnea, and elevated inflammatory markers.

However, in many cases, these effusions are not infectious but rather represent a reactive inflammatory response, potentially triggered by manipulation of the parietal pleura or irritation caused by the Nuss bar. Imaging alone often fails to differentiate between sterile and infectious effusions, making pleural fluid aspiration and culture the diagnostic gold standard. In patients unresponsive to antibiotics and with negative cultures, corticosteroid therapy proved to be effective, supporting a non-infectious, inflammatory etiology in select cases.[17, 18]

Based on these findings, we developed and implemented a structured diagnostic and treatment protocol, including routine postoperative chest X-rays at three weeks and early consideration of corticosteroids in culture-negative, non-resolving effusions. Future research should aim to better understand the role of tissue manipulation, particularly from cryoanalgesia and sternal elevation, in the development of inflammatory complications such as pleural effusion, and to validate our management strategy in larger prospective cohorts.

In **Chapter 9**, we investigated postoperative infections following the Nuss procedure, identifying an overall infection rate of 4.0%. These infections showed a temporal shift from early- to late-onset over time, with Cutibacterium acnes (CA), a low-virulence skin bacterium, becoming increasingly prevalent.

The exact cause of the rising incidence of CA in late-onset Nuss bar infections remains uncertain. This trend may reflect a true increase in CA-related infections, a relative decline in other pathogens, or improved detection methods resulting from more frequent and targeted culturing. Further investigation is warranted, particularly through microbiological analysis of explanted bars, to determine whether CA acts as a true pathogen in these cases or merely represents a contaminant. The increased recognition of CA also highlights the importance of improved fixation techniques and optimal stabilizer placement to minimize mechanical instability. Bar dislocation and stabilizer loosening alter the local tissue environment through micromotion, which may facilitate colonization by low-virulence organisms such as CA.

Over the past 25 years, a wide variety of antibiotic regimens have been employed in managing Nuss bar infections, often based on individual surgeon preference.[19, 20] Based on our findings, we propose a structured, multidisciplinary approach to the diagnosis and treatment of these infections. It is equally important to remain vigilant

for non-infectious causes of postoperative erythema, such as metal hypersensitivity and terra firma-forme dermatosis.[21] The latter, though benign and easily treated with alcohol wipes, may be misinterpreted as infection or allergy, leading to unnecessary interventions.

Looking ahead, further efforts should be directed toward validating our proposed diagnostic and therapeutic protocol in larger prospective cohorts. Simultaneously, optimizing bar fixation techniques will be critical in reducing infection rates and improving long-term outcomes.

Beyond the complications mentioned in **Chapter 8 and 9**, our clinical experience has revealed a number of unusual postoperative skin changes with unclear etiology. We hypothesize that these may be related to the introduction of cryoanalgesia, specifically the effects of temporary intercostal nerve blockade on local skin physiology, leading to atypical appearances. Since these phenomena remain poorly understood at present, they underscore the need for systematic observation and documentation. Future studies should aim to elucidate their underlying mechanisms and clinical significance.

CROSS-CUTTING PERSPECTIVES

In **Part III**, we reported an evaluation of the impact of surgical wait times on treatment withdrawal among pediatric patients, identifying key predictors of withdrawal and underscoring the importance of a patient-centered approach in treatment decision-making.

In **Chapter 10**, we investigated the impact of prolonged waiting times on patients listed for pectus surgery. After an average delay of nearly five years, only 40.2% of patients still wished to undergo surgery. Withdrawal was most common in younger, asymptomatic patients and was associated with psychosocial improvement, body acceptance, and strength training or weight gain.

Interestingly, withdrawal often reflected personal growth rather than dissatisfaction with care. Many patients initially sought treatment for cosmetic or emotional reasons but later reconsidered surgery as their self-image improved. This trend suggests that psychological support and time for natural development can reduce the perceived need for surgery, especially in patients with PC. Structured strength training may help PC patients, especially those who are lean or in early adolescence.[22,23] Given that 70% of patients listed for Ravitch surgery eventually withdrew, surgical indications for PC should be reassessed, favoring non-invasive strategies when appropriate.

For patients with PE, caution is advised regarding reassessing indications for surgery. Although asymptomatic, many patients exhibit subclinical cardiopulmonary impairment that only becomes evident through improvement following surgical correction.[24] This group of patients, although asymptomatic does benefit from surgery.

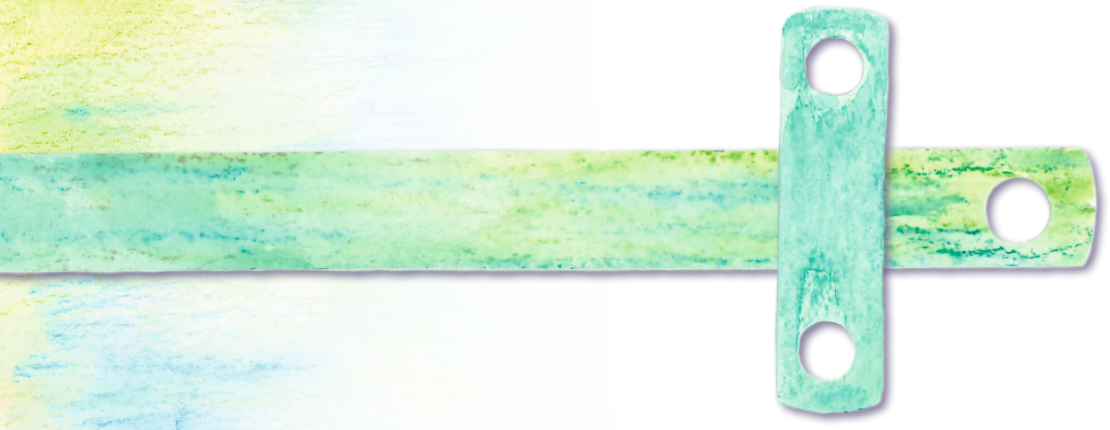
Additionally, practical barriers also emerged as a reason for withdrawal, such as changes in life circumstances making surgery less feasible. Delays in surgery may result in missed opportunities or increased technical difficulty due to chest wall rigidity with age.[25] Thus, timing remains crucial, and early support, both physical and psychological, should be part of the treatment pathway.

Future efforts should include developing tools to help predict who will benefit most from surgery, integrating psychological counseling early in care, and promoting non-invasive therapies. A nuanced, individualized approach to timing and type of intervention will likely improve long-term outcomes and patient satisfaction.

REFERENCES

1. Van Braak H, de Beer SA, Twisk JWR, et al. Improving Quality of Life With Dynamic Compression Bracing in Patients With Pectus Carinatum. J Pediatr Surg. 2025;60(1):161975.
2. De Beer S, Volcklandt S, de Jong J, et al. Dynamic Compression Therapy for Pectus Carinatum in Children and Adolescents: Factors for Success. J Pediatr Surg. 2023;58(8):1440-5.
3. Abramson H, Aragone X, Blanco JB, Ciano A, Abramson L. Minimally invasive repair of pectus carinatum and how to deal with complications. J Vis Surg. 2016;2:64.
4. Port E, Hebal F, Hunter CJ, Malas B, Reynolds M. Measuring the impact of brace intervention on pediatric pectus carinatum using white light scanning. J Pediatr Surg. 2018;53(12):2491-4.
5. Szafer D, Taylor JS, Pei A, et al. A Simplified Method for Three-Dimensional Optical Imaging and Measurement of Patients with Chest Wall Deformities. J Laparoendosc Adv Surg Tech A. 2019;29(2):267-71.
6. Wong KE, Gorton GE 3rd, Tashjian DB, Tirabassi MV, Moriarty KP. Evaluation of the treatment of pectus carinatum with compressive orthotic bracing using three dimensional body scans. J Pediatr Surg. 2014;49(6):924-7.
7. Song SH, Kim CH, Moon DH, Lee S. Usefulness of 3-Dimensional Body Surface Scanning in the Evaluation of Patients with Pectus Carinatum. Korean J Thorac Cardiovasc Surg. 2020;53(5):301-5.
8. Taylor JS, Madhavan S, Szafer D, et al. Three-Dimensional Optical Imaging for Pectus Excavatum Assessment. Ann Thorac Surg. 2019;108(4):1065-71.
9. Haecker FM, Sesia S. Non-surgical treatment of pectus excavatum. J Vis Surg. 2016;2:63.
10. Obermeyer RJ, Cohen NS, Kelly RE, et al. Nonoperative management of pectus excavatum with vacuum bell therapy: A single center study. J Pediatr Surg. 2018;53(6):1221-5.
11. Patel AJ, Hunt I. Is vacuum bell therapy effective in the correction of pectus excavatum? Interact CardioVasc Thorac Surg. 2019;29(2):287-90.
12. Zacha S, Lopez M, Bilas J, et al. One- versus Two-Minute Intercostal Nerve Cryoanalgesia in Children Undergoing Surgery for Funnel Chest Deformity. J Pers Med. 2024;14(8).
13. Zeineddin S, Goldstein SD, Linton S, et al. Effectiveness of one minute per level intercostal nerve cryoablation for postoperative analgesia after surgical correction of pectus excavatum. J Pediatr Surg. 2023;58(1):34-40.
14. Mariconti S, Bronco A, Pellicioli I, et al. Earlier preoperative percutaneous intercostal cryoanalgesia improves recovery after pectus excavatum surgery. Reg Anesth Pain Med. 2024.
15. Velayos M, Alonso M, Delgado-Miguel C, et al. Percutaneous Cryoanalgesia: A New Strategy for Pain Management in Pectus Excavatum Surgery. Eur J Pediatr Surg. 2022;32(1):73-9.
16. Velayos M, Alonso M, Estefanía K, et al. Ultrasound-Guided Percutaneous Cryoanalgesia for Pectus Excavatum: When Should It be Applied? Eur J Pediatr Surg. 2023;33(1):61-7.
17. Cheng YL, Lin CT, Wang HB, Chang H. Pleural effusion complicating after Nuss procedure for pectus excavatum. Ann Thorac Cardiovasc Surg. 2014;20(1):6-11.
18. Castellani C, Schalamon J, Saxena AK, Höellwarth ME. Early complications of the Nuss procedure for pectus excavatum: a prospective study. Pediatr Surg Int. 2008;24(6):659-66.
19. Shin S, Goretsky MJ, Kelly RE Jr., Gustin T, Nuss D. Infectious complications after the Nuss repair in a series of 863 patients. J Pediatr Surg. 2007;42(1):87-92.

20. Calkins CM, Shew SB, Sharp RJ, et al. Management of postoperative infections after the minimally invasive pectus excavatum repair. J Pediatr Surg. 2005;40(6):1004-7; discussion 7-8.
21. Pitt JB, Carter M, Zeineddin S, et al. Chest Wall Dermatitis Patterns Following Thoracoscopic Intercostal Nerve Cryoablation for Surgical Correction of Pectus Excavatum. J Pediatr Surg. 2024;59(9):1687-93.
22. Kuru P, Cakiroglu A, Er A, et al. Pectus Excavatum and Pectus Carinatum: Associated Conditions, Family History, and Postoperative Patient Satisfaction. Korean J Thorac Cardiovasc Surg. 2016;49(1):29-34.
23. Van Braak H, de Beer SA, Zwaveling S, et al. Evaluating Inter- and Intraobserver Agreement on Pectus Carinatum Severity and Treatment Outcomes: A Comparison of Subjective and Objective Assessment Methods. Eur J Pediatr Surg. 2025;35(3):232-9.
24. Raggio IM, Toselli L, Valle M, et al. Cardiac decompression and right ventricular function improvement after bar removal in patients with pectus excavatum. Int J Cardiovasc Imaging. 2024;40(11):2383-90.
25. Coughlin AC, Ahsanuddin S, Inglesby D, et al. When to Nuss? patient age as a risk factor for complications of minimally invasive repair of pectus excavatum: a systematic review and meta-analysis. Pediatr Surg Int. 2022;38(3):365-75.



Chapter 12

Nederlandse samenvatting

SAMENVATTING

Pectus carinatum (PC) en pectus excavatum (PE) zijn borstkastafwijkingen die zowel conservatief als chirurgisch succesvol behandeld kunnen worden, met in beide gevallen goede langetermijnuitkomsten. **Deel I** van deze thesis richt zich op PC. Daarin worden Dynamic Compression System (DCS)-bracing en de Ravitch operatie met elkaar vergeleken, waaruit blijkt dat DCS-bracing aanzienlijke voordelen heeft ten opzichte van opereren, en de voorkeursbehandeling in de eerste lijn zou moeten zijn. Daarnaast wordt beschreven dat DCS-bracing een positieve invloed heeft op de kwaliteit van leven van patiënten. Ook laat het onderzoek beschreven in dit deel de noodzaak van objectieve uitkomstmaten zien en worden mogelijkheden van 3D-scannen voor de diagnose van PC en het monitoren van de behandeling verkend. **Deel II** gaat over PE. Daarin wordt de effectiviteit van vacuümbel (VB) therapie beoordeeld, worden pijnbestrijdingsstrategieën na de Nuss-procedure vergeleken en worden praktische handvatten gegeven voor het omgaan met postoperatieve complicaties zoals pleuravocht, postoperatieve infecties en allergische reacties. **Deel III** beschrijft een overkoepelend onderzoek naar de invloed van wachttijden voor chirurgie op de keuze om wel of geen operatie te ondergaan, identificeert belangrijke voorspellers van uitval en onderstreept de invloed van psychologische factoren op de besluitvorming rond de behandeling.

DEEL I: PECTUS CARINATUM

In **Hoofdstuk 2** werden de uitkomsten van DCS-bracing en de Ravitch operatie bij 738 patiënten met pectus carinatum over een periode van tien jaar vergeleken. De resultaten laten zien dat DCS-bracing de voorkeur verdient als eerstelijnsbehandeling vanwege het niet-invasieve karakter, het hoge slagingspercentage en het lagere complicatierisico ten opzichte van chirurgie. Een hoge initiële compressiedruk en syndromale aandoeningen, zoals het syndroom van Marfan of Poland, zijn geassocieerd met slechtere uitkomsten. Ondanks de goede resultaten blijft onvoldoende therapietrouw een belangrijke uitdaging bij DCS-bracing. Chirurgische correctie, met name de Ravitch operatie, blijft een waardevol alternatief bij patiënten met een ongunstig profiel voor DCS-bracing.

Los van de vraag in hoeverre DCS-bracing PC kan corrigeren, werd ook onderzocht of de behandeling inderdaad de kwaliteit van leven verbetert. In **Hoofdstuk 3** rapporteren we de impact van DCS-bracing op de kwaliteit van leven bij 225 PC-patiënten van 10 tot 21 jaar. De resultaten tonen significante verbeteringen in zelf gerapporteerde lichamelijke klachten, pijn, psychische gezondheid, zelfvertrouwen en algehele kwaliteit van leven op meerdere gevalideerde vragenlijsten, met name in de eerste zes tot twaalf maanden van de behandeling. Bij patiënten met een onsuccesvolle behandeling werd

geen verbetering gezien. Bijna 90% zou opnieuw voor bracing kiezen, en 95% was tevreden met de behandeling.

Ondanks de hoge tevredenheid bemoeilijkt het ontbreken van gestandaardiseerde, objectieve eindpunten de DCS-bracebehandeling. In **Hoofdstuk 4** bespreken we de inter- en intraobservervariabiliteit bij de visuele beoordeling van PC aan de hand van gestandaardiseerde foto's van 201 patiënten voor en na behandeling. Zowel chirurgen als leeftijdsgenoten van patiënten beoordeelden esthetische uitkomsten, ernst en symmetrie; ze deden dat niet op een consistente manier en hadden slechts geringe tot matige overeenstemming. Afwijkingen gecorrigeerd door Ravitch chirurgie werden als ernstiger beoordeeld, maar leverden wel betere esthetische resultaten op. Leeftijdsgenoten beoordeelden littekens negatiever dan chirurgen. Er werd geen verband gevonden tussen uitkomsten en leeftijd of behandelingsduur. Deze bevindingen onderstrepen de beperkingen van subjectieve beoordeling en pleiten voor het ontwikkelen van objectieve meetinstrumenten om behandelresultaten te evalueren.

In reactie op de behoefte aan objectieve meetmethoden werd de rol van 3D body imaging bij klinische besluitvorming onderzocht in **Hoofdstuk 5**. In een cross-sectionele studie werd de External Haller Index (EHI) van 43 patiënten met PC vergeleken met die van 50 gezonde adolescenten. Hoewel de EHI bij patiënten met PC significant lager was, beperkte de aanzienlijke overlap met gezonde leeftijdsgenoten de bruikbaarheid als strikte diagnostische afkapwaarde. De waargenomen correlatie tussen EHI en Body Mass Index (BMI) suggereert dat niet-gecorrigeerde EHI-waarden op zichzelf onvoldoende zijn om de ernst van de afwijking of de behandelvoortgang betrouwbaar te volgen; deze correlatie onderstreept de noodzaak van BMI-gecorrigeerde referentiewaarden en verdere validatie van 3D body imaging parameters.

DEEL II: PECTUS EXCAVATUM

In **Hoofdstuk 6** werden de langetermijnuitkomsten en factoren die het succes van vacuümbeltherapie (VB) beïnvloeden onderzocht bij 259 patiënten (<18 jaar) met PE. Onder degenen die de behandeling voltooiden, was het slagingspercentage 52,1%. Een hoger slagingspercentage was geassocieerd met een langere totale behandelduur, langere dagelijkse draagduur en nachtelijk dragen. Complicaties waren minimaal en een recidief kwam zelden voor. Opvallend was dat 26,7% van de patiënten die tijdens het wachten op een Nuss-procedure werd behandeld met VB-therapie, uiteindelijk geen operatie meer nodig had. Bij vrouwelijke patiënten vormde borstontwikkeling een belangrijke belemmering; 39,3% staakte om die reden de behandeling. Bij deze patiënten kan beter vroeg gestart worden met de behandeling, of juist een afwachtend beleid gevoerd worden. Diepere afwijkingen, een flexibele borstkast en een symptomatische

presentatie waren geassocieerd met een lager slagingspercentage. Deze bevindingen benadrukken de noodzaak van een op de patiënt afgestemde behandeling om de uitkomsten van VB-therapie te verbeteren.

De meerderheid van de patiënten met PE wordt behandeld middels de Nuss-procedure. Voor de introductie van cryoablatie ging deze minimaal invasieve ingreep gepaard met aanzienlijke postoperatieve pijn, wat vaak leidde tot langere ziekenhuisopnames en een hoog opioïdgebruik. In **Hoofdstuk 7** werd de effectiviteit van cryoablatie vergeleken met continue epidurale analgesie (CEA) bij 66 kinderen die een Nuss-procedure ondergingen wegens PE. De resultaten toonden aan dat cryoablatie leidde tot significant lagere postoperatieve pijnscores, kortere opnameduur, en minder opioïdgebruik, zowel bij ontslag als één week postoperatie, in vergelijking met CEA. Hieruit kan geconcludeerd worden dat cryoablatie de voorkeursmethode voor pijnbestrijding is. Dit onderzoek laat ook zien dat routinematig profylactisch gebruik van gabapentine slechts een beperkte meerwaarde heeft bij patiënten die cryoablatie krijgen.

Hoewel de Nuss-procedure doorgaans goede resultaten geeft, ontstaan er bij deze procedure ook vaak complicaties; één daarvan is postoperatief pleuravocht, een weinig begrepen en vaak verkeerd gediagnosticeerde complicatie. In **Hoofdstuk 8** worden drie illustratieve casussen van pleuravocht na de Nuss-procedure beschreven. Is er geen Nuss-barinfectie of -dislocatie, dan ontstaat het pleuravocht waarschijnlijk door een reactieve ontstekingsrespons. Deze kan zich klinisch presenteren als, en op beeldvorming lijken op, een longontsteking en reageert vaak goed op behandeling met corticosteroïden. De drie casussen onderstrepen het belang van diagnostische pleurapuncties om de juiste diagnose te kunnen stellen. Op basis van de opgedane ervaring met deze complicatie werd een protocol voor diagnostiek en behandeling voorgesteld om clinici te helpen deze complicatie effectiever te herkennen en te behandelen.

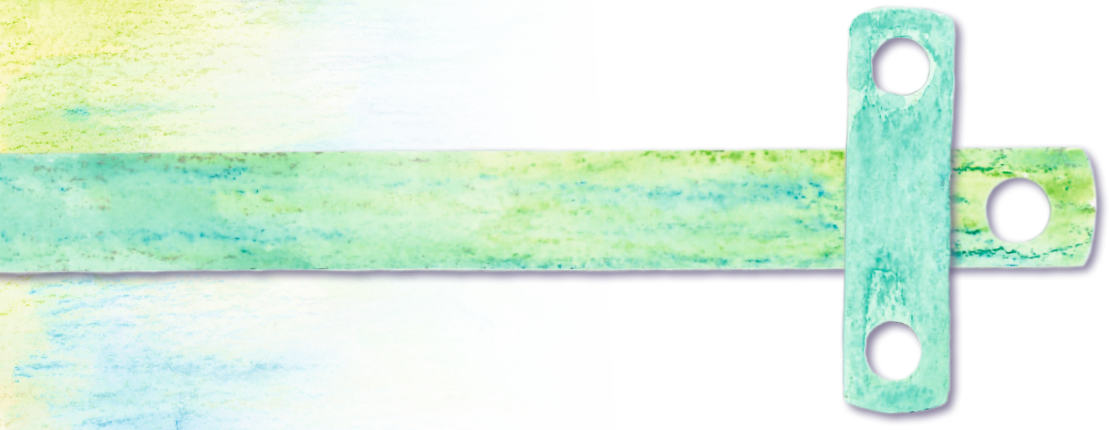
Een andere complicatie van de Nuss-procedure is postoperatieve infecties. In **Hoofdstuk 9** werden incidentie, kenmerken en behandeling van Nuss-barinfecties geanalyseerd bij 695 patiënten die tussen 1999 en 2024 werden behandeld. Van alle patiënten ontwikkelde 4,0% een postoperatieve infectie. Infecties presenteerden zich doorgaans met erytheem, pijn en exsudaat. In de loop van jaren verschoof het patroon van vroege naar late infecties. De meeste infecties waren diepe infecties, waarvoor chirurgisch ingrijpen nodig was. Oppervlakkige infecties werden behandeld met antibiotica. Dislocatie van de Nuss bar en luxatie van de stabilisatieplaat waren belangrijke risicofactoren voor infectie. Daarnaast waren infecties geassocieerd met een langere procedure bij het uithalen van de bar. Op basis van de bevindingen is een

gestructureerd protocol voor diagnostiek en behandeling ontwikkeld. De aanbeveling is om acute infecties te behandelen met zes weken flucloxacilline plus rifampicine oraal, en laaggradige infecties langdurig (tot aan de verwijdering van de Nuss-bar) met amoxicilline of clindamycine. Bij patiënten met persisterend erytheem zonder systemische verschijnselen of afwijkende beeldvorming moet aan een implantaatallergie worden gedacht. Daarnaast werd in dit hoofdstuk terra firma-forme dermatosis uitgelicht als een goedaardige postoperatieve huidaandoening die eenvoudig met alcoholdoekjes te verwijderen is.

DEEL III: OVERKOEPELENDE PERSPECTIEVEN

Hoofdstuk 10 rapporteert onderzoek naar de impact van langdurige wachttijden (in dit geval grotendeels als gevolg van de COVID-19-pandemie) op de beslissing om al dan niet een operatie voor pectusafwijkingen te ondergaan. In een cohort van 107 patiënten wilde na een gemiddelde wachttijd van bijna vijf jaar nog slechts 40,2% van de patiënten geopereerd worden. Het uitvalpercentage verschilde tussen PE-, PC- en flaringpatiënten. De belangrijkste reden om af te zien van operatie was bij PE toegenomen lichaamsacceptatie, en bij PC en flaring lichamelijke ontwikkeling (krachttraining of gewichtstoename). Patiënten die afzagen van operatie rapporteerden een grotere afname van zowel fysieke als psychosociale klachten dan degenen die op de wachtlijst bleven. Lagere leeftijd en het ontbreken van lichamelijke klachten bij plaatsing op de wachtlijst waren significante voorspellers van uitval. Conservatieve behandeling was tijdens de wachttijd slechts bij een minderheid succesvol. Deze bevindingen benadrukken het belang van vroege psychologische begeleiding en fysieke training als onderdeel van de standaardzorg voor jonge, asymptomatische patiënten, waardoor chirurgie in deze gevallen waarschijnlijk vermeden kan worden.

Meer weten over pectusafwijkingen? Luister dan ook eens deze podcast.



Appendices

List of publications

PhD portfolio

Dankwoord

Curriculum vitae

LIST OF PUBLICATIONS

Publications in this thesis

2025 Van Braak H, Houter E, de Beer SA et al. Differences in Chest Wall Between Dutch Adolescents with Pectus Carinatum and the General Pediatric Population: A Three-Dimensional Scanning Analysis. J Pediatr Surg. 2025. ***Online ahead of print***

2025 Van Braak H, Terheggen-Lagro SWJ, Israels J et al. Diagnostic and Therapeutic Challenges of Pleural Effusion After the Nuss Procedure: A Case Series. J Pediatr Surg Case Rep. 2025;121:103085.

2025 Van Braak H, de Beer SA, Oomen MWN, et al. Time Heals: The Impact of Waiting Times on Pediatric Patients' Decisions to Decline Pectus Surgery. J Pediatr Surg. 2025;60(11):162504.

2025 Van Braak H, de Beer SA, Zwaveling S, et al. Evaluating Inter- and Intraobserver Agreement on Pectus Carinatum Severity and Treatment Outcomes: A Comparison of Subjective and Objective Assessment Methods. Eur J Pediatr Surg. 2025;35(3):232-239.

2025 Van Braak H, de Beer SA, al Ghouch Y, et al. 15 years of vacuum bell therapy for pectus excavatum: long-term outcomes and influencing factors. J Pediatr Surg. 2025;60(2):161891.

2025 Van Braak H, de Beer SA, Twisk JWR, et al. Improving Quality of Life with Dynamic Compression Bracing in Patients with Pectus Carinatum. J Pediatr Surg. 2025;60(1):161975.

2024 Van Braak H, de Beer SA, de Jong JR, et al. Intercostal Nerve Cryoablation or Epidural Analgesia for Multimodal Pain Management after the Nuss Procedure: A Cohort Study. Eur J Pediatr Surg. 2024;34(6):488-492.

2024 Van Braak H, de Beer SA, Zwaveling S, Oomen MWN, de Jong JR. Ravitch Surgery or Dynamic Compression Bracing for Pectus Carinatum: A Retrospective Cohort Study. Ann Thorac Surg. 2024;117(1):144-150.

Submitted for publication

2025 Van Braak H, van Elzakker EPM, de Beer SA et al. Nuss Bar Infections: Risk Factors and Management Strategies.

Other publications

2025 Docter D, van Braak H, de Jong B, et al. Pediatric External Hemorrhoids: Clinical Characteristics and Outcomes of Conservative Treatment Versus Injection Sclerotherapy. Eur J Pediatr. 2025;184:552.

2023 Van Braak H, Gorter RR, van Wijk MP, de Jong JR. Laparoscopic Roux-en-Y feeding jejunostomy as a long-term solution for severe feeding problems in children. Eur J Pediatr. 2023;182(2):601-607.

PHD PORTFOLIO

PhD student	Hendrik van Braak
Primary thesis advisor	Prof. dr. L.W.E. van Heurn
Other thesis advisor	Dr. J. R. de Jong
Research programme	10404 Orthopedics, Trauma Surgery and Rehabilitation
Title of thesis	Advancing Pectus Deformity Care: Evaluation of Current Treatments, Complications and Future Innovations

Courses	**Year**
Leiden University Onboarding Programme Inform & Connect	2024
Responsible Research	2024
eBROK + eBROK verdiepingscursus WMO	2024
Basic Methods and Reasoning in Biostatistics	2025
Academic Writing for PhDs	2025
Career planning for PhDs and Postdocs	2025
Seminars	
Monthly research meetings, department of pediatric surgery AUMC & department of surgery LUMC	2024-2025
(Inter)national congresses and presentations	
Wetenschappelijke vergadering Nederlandse Vereniging voor Kinderchirurgie	2024
Oral presentation – ***Ravitch surgery or dynamic compression bracing for pectus carinatum: a retrospective cohort study***	
Award – ***Ganzenveer for best pediatric surgical paper of 2023***	
European Pediatric Surgeons' Association (EUPSA) Congress Bologna	2024
Wetenschapsdag Chirurgie Regio A	2024
Oral presentation – ***15 years of vacuum bell therapy for pectus excavatum: long-term outcomes and influencing factors***	
Annual Congress of the Chest Wall International Group and Phoenix Advanced Pectus Course	2025
Oral presentation – ***Improving quality of life with dynamic compression bracing in patients with pectus carinatum***	
Oral presentation – ***15 years of vacuum bell therapy for pectus excavatum: long-term outcomes and influencing factors***	

European Pediatric Surgeons' Association (EUPSA) Congress Dubrovnik	2025
Poster presentation – ***Nuss bar infections: risk factors and management strategies***	
Poster presentation – ***Pleural effusion after Nuss procedure: a case series***	
Annual Congress of the Chest Wall International Group and Japan Advanced Pectus Course	2025
Oral presentation – ***Nuss bar infections: risk factors and management strategies***	
Oral presentation – ***Time heals: the impact of waiting times on pediatric patients' decisions to decline pectus surgery***	
Poster presentation – ***Pleural effusion after Nuss procedure: a case series***	
European Respiratory Society Congress Amsterdam	2025
Poster presentation – ***Pleural effusion after Nuss procedure: a case series***	
Other	
Student projects coordinator, department of pediatric surgery	2024-2025
Member Scientific Committee Chest Wall International Group	2025
Teaching	
Lecturing	
Pediatric basics – dehydration	2025
Pediatric basics – growth and development	2025
Pediatric basics – how to make contact with children?	2025
Supervising	
Master's thesis (E. Houter)	2024-2025
Master's thesis (F. Nasra)	2024-2025
Master's thesis (F.E. Fabels)	2025
Master's thesis (Q.L.M. Aker)	2025

DANKWOORD

Dit proefschrift is het resultaat van een leerzame periode, die ik niet had kunnen voltooien zonder de hulp van velen, waarvan ik er hieronder een aantal in het bijzonder wil bedanken. Allereerst wil ik hierbij mijn waardering uitspreken voor alle patiënten en hun ouders die bereid waren deel te nemen aan de studies.

Beste prof. dr. L.W.E. Van Heurn, beste Ernest, hartelijk dank dat je mijn promotor wilde zijn. Hartelijk dank voor de kans die ik kreeg om dit traject in te gaan en voor het gladstrijken van de vele logistieke hobbels die we afgelopen jaar moesten nemen om dit promotietraject mogelijk te maken. Daarnaast nam je altijd uitgebreid de tijd om manuscripten met mij door te nemen en carrièregesprekken te voeren, hartelijk dank daarvoor!

Beste dr. J.R. de Jong, beste Justin, zonder jou was ik niet geweest waar ik nu sta. Van meekijken op de operatiekamer in klas zes, via een bachelorscriptie naar een promotie. Wie had dat gedacht. Hartelijk dank voor je fijne begeleiding, je niet aflatende enthousiasme, positiviteit, Geloof en het vertrouwen wat je mij altijd gaf. Ik heb veel van je geleerd, niet alleen als arts maar ook als mens, en hoop dat komende jaren te blijven doen!

Niet te vergeten de andere leden van het pectusteam van het Amsterdam UMC: dr. S.A. de Beer, dr. M.W.N. Oomen en dr. S. Zwaveling. Beste Sjoerd, leuk dat ik het stokje van jou over mocht nemen. Dank voor je aanstekelijke enthousiasme, je eindeloze stroom aan ideeën, en de gezellige momenten tijdens congressen en op de OK in Leiden. Je hebt mij niet alleen veel geleerd over de pectuschirurgie, maar ook wegwijs gemaakt in de community eromheen. Beste Matthijs, bedankt voor alle keren dat ik mee mocht doen op OK in Leiden, het meelezen én -schrijven van de manuscripten en voor je hulp bij het opzetten van het POLAR-project. Beste Sander, ook jij ontzettend bedankt voor je betrokkenheid, de samenwerking op de OK en je scherpe blik bij het meelezen en -schrijven van manuscripten.

Dan de andere kinderchirurgen van het Amsterdam UMC, en in het bijzonder prof. dr. J.P.M. Derikx. Beste Joep, hartelijk dank voor het meedenken over mijn onderzoekstraject én over onderzoek in bredere zin. Heel fijn dat je altijd zo toegankelijk en benaderbaar was! Ook wil ik de chirurgen in het Leiden UMC, en in het bijzonder dr. W.J. van der Made bedanken. Beste Wendeline, bedankt voor het warme welkom in Leiden en voor de keren dat ik met jou mee mocht opereren!

Alle onderzoekers van de kinderchirurgie, Lore, Fien, Sophie, Malou, Daniël, Charlotte, Anne, Said, Maaike, Anne, Daniël en Adinda, hartelijk dank voor het warme bad waarin ik terecht kwam toen ik mijn promotietraject begon. Ik zal mijn allereerste congres en aansluitende vakantie in de Toscaanse villa, het project Kerstkaarten, het jaar in de Bunker en de (escalatie)borrels niet snel vergeten. Jullie zijn geweldige collega's, ik ga jullie missen! Sibbeliene, Heleen, Mirthe, Lindsey, Nadia, Ryan, Tessa, Priscilla en Eelke, ook zonder jullie was dit traject ongetwijfeld een stuk stroperiger en minder gezellig verlopen!

De Leidse promovendi van het Greenlight Team, in het bijzonder Stefan, Roderick, Mo en Tom, dankjulliewel voor de gezellige tijd, al was het kort, in de bungalow.

Ook alle collega's van het Onze Lieve Vrouwen Gasthuis, dankjulliewel voor de warme ontvangst, voor alles wat ik van jullie mag en kan leren, en voor de gezelligheid tijdens en na het werk!

Dr. M. Gordjestani, bedankt voor het beschikbaar stellen van de 3D scanner. Dr. W.J. van der Schoor, bedankt voor het faciliteren van het onderzoek op de Jacobus Fruytier Scholengemeenschap!

Vrienden en familie, bedankt voor alle steun en betrokkenheid tijdens het promotietraject. In het bijzonder Derian en Rutger, dank voor de spontane escalaties en de vele (wilde) avonturen die we samen hebben beleefd. De avonden, weekenden en vakanties met jullie waren echt een uitlaatklep voor mij, en dat hebben jullie geweten! Daniël, Timon en Gerard, dankjewel voor jullie vriendschap en het luisterend oor op momenten dat het nodig was. Erik dankjewel dat je mijn paranimf wil zijn, mooi om samen met jou de medische wereld te ontdekken. Mama dankjewel voor de prachtige omslag van dit proefschrift. Papa, Marije, Gerrit, Pieternel, Henri, Rinske, Willem Jan, Reinier, Aren, Elsabeth en Machteld, dank voor alle hulp en gezelligheid in de afgelopen periode bij alles wat er naast het promotietraject speelde. Daniëlle, ook jij bedankt voor je geduld, steun en liefde tijdens mijn promotieonderzoek.

En dan als laatste, speciale dank aan Labrinus, jij was het die mij enthousiast maakte voor de geneeskunde en mij prikkelde om wetenschappelijk onderzoek te gaan doen. Bedankt voor onze mooie avonturen (samen met Adrianus), voor alles wat ik van je geleerd heb, dat ik je paranimf mocht zijn, en nog veel meer. Je bent en blijft een voorbeeld voor mij.

CURRICULUM VITAE

Hendrik (Dirk) van Braak werd op 17 februari 1997 geboren te Veenendaal. Na het volgen van tweetalig onderwijs aan de Jacobus Fruytier Scholengemeenschap en het behalen van zijn gymnasiumdiploma in 2015, begon hij in 2016 met de studie Geneeskunde aan de Universiteit van Amsterdam. Tijdens de wachttijd van zijn coschappen begon hij zijn wetenschapsstage bij de Kinderchirurgie onder leiding van Dr. J.R. de Jong, wat leidde tot een eerste publicatie. Tijdens deze stage werd zijn interesse voor de wetenschap aangewakkerd, waarna hij gedurende de coschappen in de avonduren wetenschappelijk onderzoek bleef doen. Begin 2024 rondde hij zijn geneeskundestudie af met een senior coschap bij de afdeling Heelkunde van het Onze Lieve Vrouwen Gasthuis Oost in Amsterdam. Het onderzoek dat hij parallel aan zijn studie uitvoerde, resulteerde in een promotietraject bij de Kinderchirurgie (bij prof. dr. L.W.E. van Heurn en dr. J.R. de Jong), waarvoor hij vanaf mei 2024 een aanstelling kreeg als arts-onderzoeker bij het Leids Universitair Medisch Centrum. Het promotietraject richt zich op het evalueren en verder ontwikkelen van de huidige behandelingen voor pectus carinatum en pectus excavatum bij kinderen. Sinds juni 2025 werkt hij als ANIOS (Arts Niet In Opleiding tot Specialist) bij de afdeling Heelkunde in het Onze Lieve Vrouwe Gasthuis in Amsterdam. Naast zijn werk bij de chirurgie beoefent hij nog steeds met veel plezier het ambacht van goudsmeden.