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Systematic review of physiological and psychological outcomes of surgery for pectus excavatum supporting commissioning of service in the UK

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ABSTRACT

Background Pectus excavatum (PEx) is the most common congenital chest wall abnormality affecting 1 in 400 births in the UK. PEx is associated with significant physiological and psychological impairment. While readily surgically correctable, the benefits that surgery can bring have been debated and proven difficult to objectively measure. In the UK, this has led to the decommissioning of PEx surgery. The aim of this review is to conduct a systematic search of the literature on PEx surgery to assess physiological and psychological outcomes.

Methods A systematic review of the MEDLINE (PubMed), Embase and Cochrane databases was performed. Articles were sought which included patients undergoing surgery for PEx and reported on changes in cardiopulmonary measures, symptoms, quality of life and psychological assessments before and after surgical repair. Last search was performed in July 2022 and relevant findings were synthesised by narrative review.

Results Fifty-one articles were included in qualitative synthesis, with 34 studies relating to physiological outcomes and 17 studies relating to psychological and quality of life measures. Twenty-one studies investigated pulmonary function at rest. There was no change in forced vital capacity or forced expiratory volume in 1 second following open repair and transient reductions followed closed repair. In the 11 studies investigating echocardiography, transthoracic rarely demonstrated cardiac compression; however, transoesophageal demonstrated intraoperative relief in cardiac compression in severe cases. Sixteen studies investigated exercise testing (cardiopulmonary exercise testing, CPET), 12 of which demonstrated significant improvement following surgery, both in maximal oxygen consumption and oxygen pulse. Seventeen studies investigated quality of life, all but one of which showed improvement following repair of PEx. All papers that reported on patient satisfaction following surgery found high rates, between 80% and 97%.

Discussion While the majority of studies to date have been small and data heterogeneous, the literature shows that for many patients with PEx, there exists a cardiopulmonary limitation that while difficult to objectify, is likely to improve with surgical repair. Resting parameters offer little yield in aiding this except in the most severe cases. CPET therefore offers a better option for dynamic assessment of this limitation and improvements following repair. Surgery significantly improves psychological well-being and quality of life for patients with PEx.

WHAT IS ALREADY KNOWN ON THIS TOPIC

⇒ The benefits of surgical repair of pectus excavatum (PEx) are debated and changes in cardiopulmonary function has been difficult to objectively measure—this has lead National Health Service UK to recently decommission surgery for PEx, affecting access to services.

WHAT THIS STUDY ADDS

⇒ This paper is an updated review of physiological and psychological outcomes following PEx surgery.

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ Cardiopulmonary exercise testing (CPET) has demonstrated improvements in exercise limitation following surgery both in adults and children however clinical correlation with patient's symptoms is necessary before minimally clinically important differences can be established.
⇒ While resting parameters are little aid to assessing all but the most severe cases of PEx, CPET can better clarify patients who gain physiological benefit from repair especially given minimally invasive surgery with lower morbidity.
⇒ There is often a large psychological component to patients experience of PEx and this should be assessed equally along with anatomical severity in shared decision making regarding surgical repair.

INTRODUCTION

Overview

Pectus excavatum (PEx), or 'funnel chest', is the most common congenital chest wall abnormality, affecting approximately 1/400–1/1000 live births.[1] It is characterised by a depression of the anterior chest wall (sternum and lower costal cartilages) which can vary from small and barely perceptible to a very large tunnel towards the spine resulting in a 'funnel' shaped thorax.[2]

Traditionally, PEx has in general been regarded as a purely cosmetic problem which has fuelled debate over the indications for surgical repair, which is the topic of this review. While for many patients with PEx, the defect is mild and well tolerated, for some it is a source of significant psychological distress. This distress most commonly occurs during teenage years, a time of crucial physical and social development and can lead to anxiety, lower self-esteem and withdrawal from social activities significantly impacting patients' quality of life (QoL). In others, PEx is associated with symptoms of exercise limitation, dyspnoea and palpitations, as displacement of the sternum disrupts their physiological reserve. Disease severity and symptom burden can vary widely and are patient-specific and not always correlated with the degree of anatomical depression.[3] While subjective complaints of PEx have been well established, finding objective measures of this has proven difficult. So far, it has remained unclear whether the basic pathophysiologic problem is primarily ventilatory or cardiovascular (or both) arising from compression of the right ventricular (RV) outflow tract by the displaced sternum.

The outcomes from surgical repair for PEx are the matter of debate and the subject of this review. Pectus surgery, whether open or particularly with introduction of closed techniques, is safe to perform in children and adults with minimal complications or patient morbidity.[4 5] Both open and closed repair provide excellent cosmetic results and greater than 80%–90% of patients are satisfied with the results in most large case series.[6] Patients frequently report subjective symptom resolution and improvement in exercise tolerance following surgical repair; however, objective measures of this have been difficult to capture with a wide variation in published results.[7 8] The existing literature is inconclusive, hampered by small, statistically underpowered patient cohorts, short-term versus long-term results, rest versus exercise studies, and inconsistent testing measures. In countries where cosmetic impact is an indication for surgery, such as the USA, there is little call to study physiological outcomes in PEx. Conversely, in the UK, this controversy around the physiological and psychological impact of PEx, has led the National Health Service (NHS) to decommission pectus surgery treatment, with far reaching consequences for access to treatment for patients with PEx.

Objectives of review

The objective of this review is to conduct a systematic search of the literature on the surgical repair of PEx to assess what improvement, if any, surgery has on physiological and psychological outcomes for patients with PEx.

METHODS

Search strategy, study identification and selection

A systematic review was performed according to the Preferred Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines 2016.[9] Articles were considered, which included patients undergoing surgical repair for PEx including both open surgery (modified Ravitch procedure) or minimally invasive repair of pectus excavatum (Nuss procedure) and reported on long-term outcomes of interest. These outcomes could be divided into physiological, namely cardiopulmonary function changes (eg, cardiopulmonary exercise testing (CPET), pulmonary function testing (PFTs), transthoracic echocardiography (TTE)) and changes in symptom burden, or psychological and change in QoL scores, pre and post-surgical intervention.

The Medline (via Pubmed), EMBASE and Cochrane databases were searched from database inception to present, with the last search undertaken in July 2022. There were no limits used and restrictions on the basis of date or language of publication at time of searching however only articles available in English were included in the full text review. The included search terms were: "Funnel Chest" [Mesh] OR "Funnel Chest" OR "Pectus Excavatum" AND "Thoracic Surgery" [Mesh] OR "Thoracic Surgical Procedures" [Mesh] OR Repair OR "Pectus Surgery" OR Nuss OR Ravitch Or "Minimally Invasive" OR "Open Surgery" OR Surgery OR "Surgical Intervention". The full search strategy, as applied to the Medline and Embase databases, is outlined in online supplemental appendix 1.

After the removal of duplicates, titles and abstracts were screened for relevance. Full-text articles were reviewed by two reviewers (JW and RW), according to the inclusion and exclusion criteria which is outlined in table 1. Reference lists of included articles and previous reviews of interest were manually searched for additional articles,

Table 1 Inclusion/exclusion criteria

Inclusion criteria	Exclusion criteria
► Randomised controlled-trials, cohort studies, case–control studies and case series including greater than five patients. ► Human studies only. ► If multiple articles had overlapping cohorts (determined by institution and year), only the most recent publication was included. ► Reports published in English language.	► Individual case reports and case series with fewer than five patients included. ► Articles describing technical aspects of surgical technique or those that purely reported on perioperative or short term outcomes. ► Animal studies. ► Publications that described surgical techniques that differ significantly from current techniques were excluded; in practice, this removed all publication prior to 1989.

which were assessed according to the same outlined inclusion and exclusion criteria. A grey literature search was carried out to find other articles of interest not identified on database searching. Disagreements were resolved by discussion. If this did not result in consensus, the third author's opinion (KR) was decisive.

Data extraction

Relevant data were collected by a single reviewer (JW) using a predefined pro forma. Data were sought on the following items from each included article including study characteristics, physical characteristics of subjects, type of surgical repair performed and outcomes of interest.

Relevant findings from all included studies were synthesised by narrative review.

Patient and public involvement

There were no patients involved in this review article.

RESULTS

Results of the search

Following the search strategy, 5464 potentially eligible papers were initially identified and an additional internet search yielded a further 2 studies. Reference management software was used to remove all duplicate references leaving 3559 studies for evaluation. These were screened by title and abstract and 3385 could be excluded leaving 174 studies for full-text review. After application of inclusion/exclusion criteria outline in table 1, 51 texts were eligible for inclusion in our review. These were classified by outcomes of interest with 34 studies relating to physiological outcomes and 17 studies relating to psychological and QoL measures. The review process is illustrated as a PRISMA flow diagram in figure 1. The study characteristics and primary outcomes are outlined in online supplemental tables 1 and 2 with diagrammatic representation of results seen in figures 2 and 3, respectively. All included articles were published between 1984 and 2022.

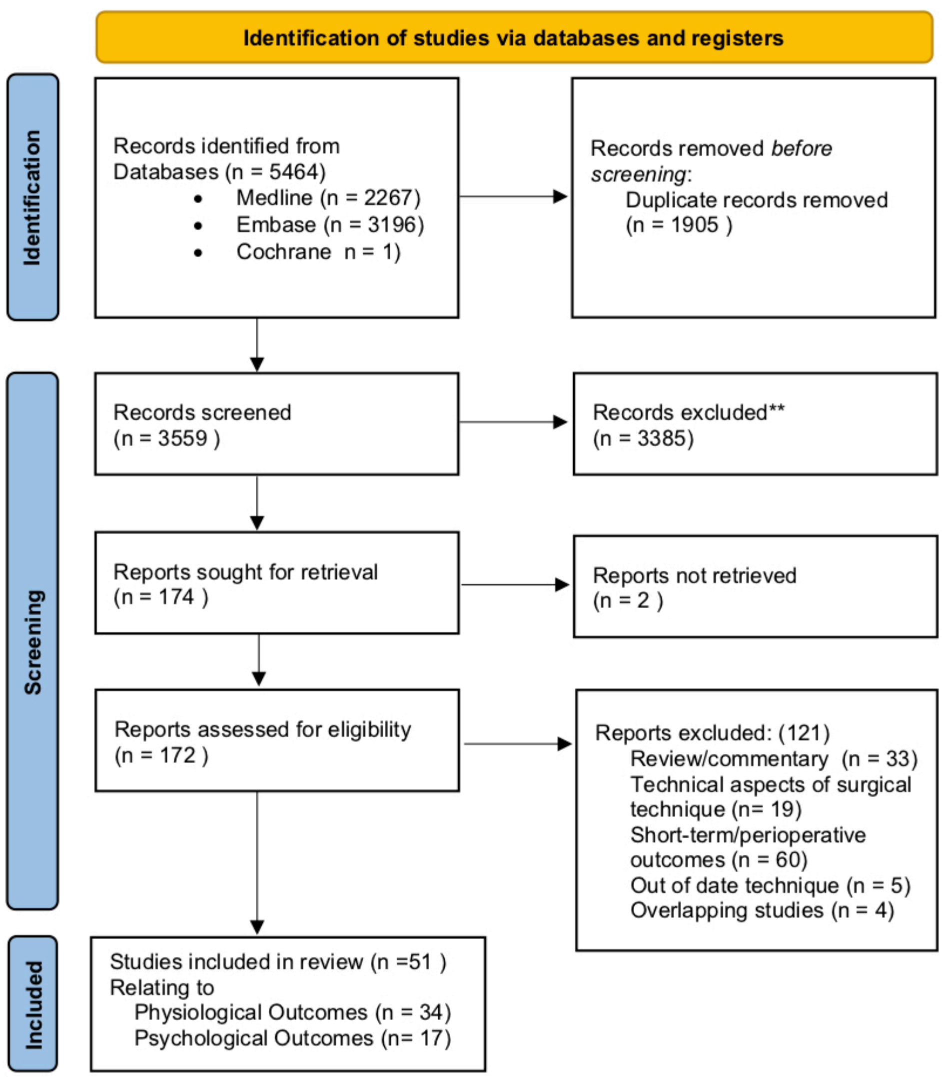

Figure 1 PRISMA 2020 flow diagram for new systematic reviews. PRISMA, Preferred Items for Systematic Reviews and Meta-Analyses. **Records excluded based on screening of titles and abstracts for relevance.

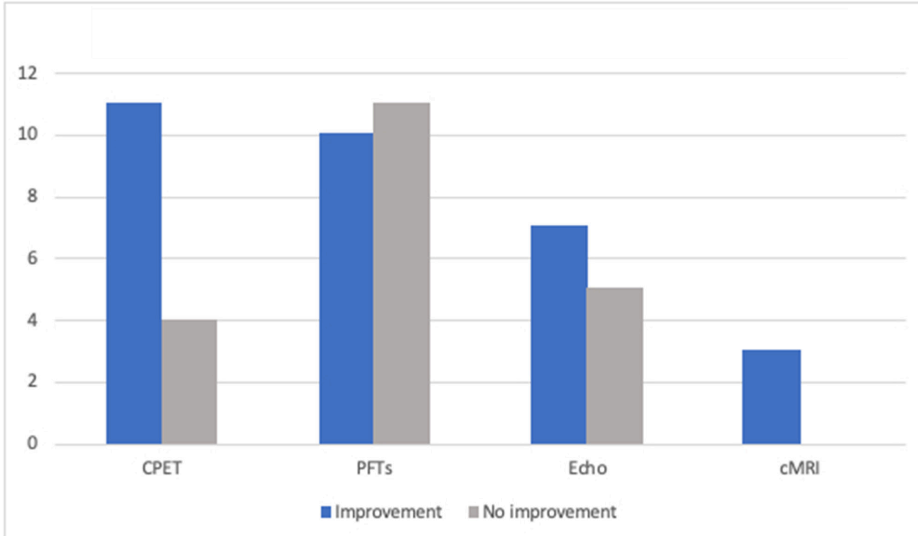

Figure 2 Studies of cardiopulmonary outcomes following pectus repair. cMRI, cardiac MRI; CPET, cardiopulmonary exercise testing; PFT, pulmonary function testing.

Cardiopulmonary and physiological outcomes

Thirty-four studies have been published which investigate cardiopulmonary and physiological outcomes of PEx surgery, the major characteristics of which are shown in online supplemental table 1. All of these consisted of either cohort studies or case series, 23 of which were carried out prospectively and ten of which retrospectively. Two studies were carried out in multiple centres.[10 11] Five of the studies used a control group, four of which used age-matched healthy controls and one used patients with PEx not undergoing surgery.[10 12–15]

Cardiopulmonary exercise testing

Sixteen studies have investigated exercise testing after surgical repair of PEx.[14–27] The primary outcome measures reported were maximal oxygen consumption (VO_{2max}), VO_{2max} per kilogram and oxygen pulse, which is a surrogate for stroke volume. Eleven of these studies were carried out in a paediatric population and five in adult populations.

In children, four of these studies included children undergoing Ravitch procedure[10 14 15 27] and seven Nuss procedure. Of those undergoing Nuss procedure, bars were still in place at the end of the follow-up period in all but two of the studies.[23 25] Seven of the 11 studies showed overall improvement in CPET variables after surgery with mean increased of VO_{2max} ranging from 6% to 40% and mean improvements in in oxygen pulse of 10%–44%.[10 12–14 18 27 28] Four studies found CPET variables to be unchanged after surgery.[15 17 20 25] Six of the studies demonstrated improvements in VO_{2max} and eight showed improvements in O_2 pulse. Two studies demonstrated improvement in O_2 pulse but not VO_{2max} after surgery.[13 20]

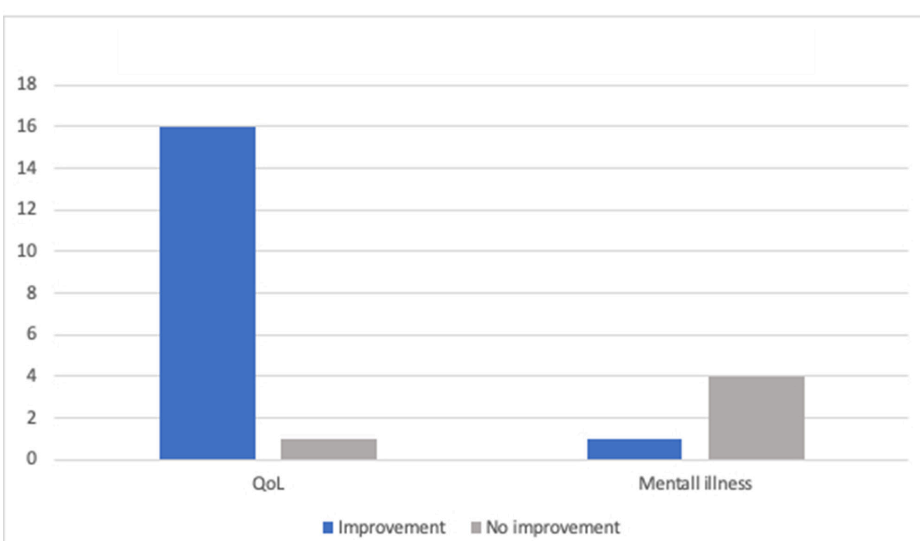

Figure 3 Studies of psychological outcomes following pectus repair. QoL, quality of life.

Of the five studies in adults, two studies[22 26] examined patients undergoing Ravitch and three studies[16 19 24] examined patients undergoing Nuss procedure. Four of the studies demonstrated improvement post surgery,[16 22 24 26] and one study did not.[19] Both studies to examine patients undergoing Ravitch demonstrated improvements in VO_{2max} and O_2 pulse postoperatively. Of the three studies to evaluate adults undergoing Nuss repair, two demonstrated significant improvements and one did not.

Pulmonary function tests

Resting pulmonary function has been extensively studied as measure of physiological improvement postsurgical repair, with mixed results. Of the 21 studies to measure PFTs, 15 have been in children[10–14 17 20 21 23 25 29–34] and 6 in adults.[19 22 24 35–37] Six studies investigated patients undergoing Ravitch repair, all of which showed no change in PFTs after surgery.[14 22 33–36]

A small prospective study by Kowalewski *et al*, which subdivided patients into moderate and severe based on anatomical severity showed an improvement in severe subgroup only.

Sixteen studies investigated PFTs in patients with PEx undergoing Nuss procedure but at different times relative to bar insertion and removal. Post initial surgery, but prior to bar removal four studies showed a decrease in pulmonary function which was transient.[23 25 29 30] In three studies no change was observed.[11 19 24] A single study saw a transient increase in pulmonary function.[11] Assessment of pulmonary function after bar removal was performed in all but three of the studies. Three studies did not show any improvement after bar removal compared with preoperative values.[11 17 25] Nine studies demonstrated improvement in pulmonary function after bar removal.[10 12 13 20 28 29 31 32 37] The majority of studies were of paediatric patients with only two studies including adults undergoing Nuss procedure.[30 37] The majority of this improvement was driven by modest gains in forced expiratory volume in 1 second (FEV1) and forced vital capacity (FVC).

Echocardiography

Of the included studies, 12 used echocardiography as an outcome measure to assess for cardiopulmonary improvement following surgery.[13 16 17 20–22 34 38–42] Eight of these used TTE presurgery and at postsurgery while four studies used intraoperative transoesophageal echocardiography (TOE). Of the eight studies to investigate TTE, three found significant improvement postoperatively.[34 38 42] Gürkan and Kowalewski both found a significant improvement in RV function (namely RV diastolic diameter and RV stroke volume index) of 47% ($p<0.05$) and 25% ($p=0.027$), respectively. However, both these studies were small and primarily enrolled severe cases where cardiac compression was seen on preoperative studies. No significant change in the RV diastolic diameter on TTE was demonstrated postsurgical repair in the majority of studies to date.[12 13 17 20 43] One study performed TTE during exercise and found 95% of 123 patients with a mean Haller index of 4.3 to demonstrate cardiac compression, 93% of which had resolved on follow-up.[42]

Four studies have used intraoperative TOE to demonstrate relief of cardiac compression during repair with similar results.[16 39 41 44] In adults, Krueger *et al* noted significant improvement in postrepair cardiac outputs that increased to 66.2% vs 58.4% and the end-diastolic RV volume that increased to a mean of 40.8 mL vs 21.7 mL preoperatively ($p<0.001$).[41] In addition, this study found the left ventricular (LV) ejection fraction to be significantly increased after surgery (58.4%±15% vs 66.2%±6%, $p<0.001$), however, this finding has not been reproduced in other studies. Also in adults patients, Chao *et al* demonstrated an increase in right atrium (15.1%), tricuspid annulus (10.9%) and RV outflow tract (6.1%) size after surgery and increase in RV cardiac output by 38% (all $p<0.0001$).[44] Similar findings were reported by Jaroszewski *et al*, with increases in RV outflow tract velocity time integral and RV stroke volume.[16] In children, Lain *et al* showed similar results with RV end diastolic diameter increasing by 5.77 mmm, right atrium diameter by 6.63 mm and tricuspid annulus by 6.02 mm (all $p<0.0001$).[39]

Other measures of cardiopulmonary function

Three studies have used cardiac MRI to assess cardiac function after surgery.[24 45 46] The Nuss procedure was performed in each study. All studies showed improvement in right heart functional parameters postoperatively with increasing RV stroke volume and RV ejection fraction at 3 months and 1-year post repair with bar still in place. Töpper *et al* also showed significant improvements in LV stroke volume and LV ejection fraction at 1-year postsurgery but this was not seen in the other studies.

Two studies of children undergoing Ravitch procedure have used radionuclide and perfusion/ventilation scintigraphy to asses cardiopulmonary function post repair.[47 48] Peterson *et al* did not show any limitation in exercise cardiac function that could be relieved by pectus repair.[47] A study by Blickman *et al* demonstrated abnormally ventilation/perfusion ratios which normalised postoperatively in the majority of patients.[48]

QoL, psychological, social and behavioural outcomes

Seventeen studies have been published which investigate QoL, psychological, social and behavioural outcomes of PEx surgery, the major characteristics of which are shown in online supplemental table 2.[49–64] Fourteen of the studies were prospective, uncontrolled cohort studies (before-and-after design).[3 49–51 54–57 59–61 65] A single one of these was multicentre. One study was of a cross-sectional design after surgery and there was a single retrospective case series.[52 63] Three studies used control groups.[52 53 66]

Mental illness

Five of the included studies specifically investigated mental illness in patients pre and post PEx repair.[53 55 59 60 66]

Of these five studies, a study by Luo *et al* was the only study to identify patients falling above the threshold for diagnosis of a mental health disorder with a significant improvement after surgery. They report the proportion of patients with mental health disorder preoperatively of 161/266 (60.5%) falling to 79/266 (29.7%) postoperatively (p<0.001).[60] A controlled study by Bahadir *et al* found no difference in scores for depression and anxiety after surgery compared with patients with PEx who did not have surgery.[66] Three studies while finding patient's preoperative scores to be in the normal range (although the lower end of normal range), that there was a significant improvement in scores postoperatively.[53 55 59] For example, in a study by Hadolt *et al*, while preoperative patient scores were within the normal range there were significant improvements in depression (p<0.05), global severity index (p<0.03) and positive symptom distress burden (p<0.05) within this range.

Patient satisfaction

All of articles that included patient satisfaction as discrete outcome separate to QoL reported very high levels of patient satisfaction with both open and closed procedures ranging from 80% to 97%.[3 50 51 54 57 58]

Quality of life

Seventeen published studies have investigated QoL in PEx patients following repair. A small pilot study by Lawson *et al* investigating the use of a disease specific, health-related QoL (HRQoL) questionnaire (Nuss Questionnaire) found significant improvements in body image, exercise capacity and frequency of being frustrated, sad, self-conscious and isolated by both patients and parents.[49] This was supported by two later prospective cohort studies finding significant improvement (p<0.001) in social function, self-esteem and a high level of satisfaction following the Nuss procedure.[50 51] Furthermore, Lam *et al* found no significant difference in HRQoL outcomes between Nuss and Ravitch procedures.[52] This improvement was shown to persist at least 4 years after bar removal (p<0.001).[51] The largest study to date to investigate QoL after PEx surgery, a multicentre study by Kelly *et al* saw a significant improvement reported by both patients and their parents in terms of body image, physical difficulties and emotional distress and social cohesion post repair. This study did not find any correlation between degree of severity and physical or psychosocial difficulties.[3]

A controlled study by Jacobsen *et al* which compared HRQoL between children post Nuss repair and healthy age-matched controls, found significant improvement in QoL measures in patients post repair to surpass their peers at the same age.[53] A later study by the same authors using generic measure of HRQoL (CHQ) to allow better comparison against age-matched healthy control group demonstrated an improvement in physical and psychosocial HRQoL after Nuss as compared with presurgery scores.[59] Bostanci *et al* found a significant improvement in both the psychosocial and physical measures postoperatively and highly significant effect on the overall QoL (all p<0.0001).[54] A smaller cohort study looking more specifically at body image perception found a significant improvement postoperatively.[55] Kim *et al* also observed a positive impact on patients' QoL and found this to be evident early post-operatively and persistent long term with no difference before and after bar removal.[56] In adults, Kuru *et al* found significant improvement in disease specific HRQoL in and psychosocial functioning 6 months after surgery.[57] A retrospective case series found 90% of patients to report improvement in general health, exercise tolerance and social interaction after surgery.[58] In contrast to these findings, a study by Bahadir *et al*, the only study to use unoperated patients with PEx as controls did not find any significant in improvement in psychosocial functioning between groups.[66] A prospective cohort study among female patients only found a significant improvement in self-perception and psychological well-being post bar removal.[61] Zuidema *et al* investigated sports activity in adolescents and found no significant change in participation in sport (67% vs 65%) 1 year after surgery, however, did find a significant decrease in physical complaints during sports activity (p<0.001).[62]

DISCUSSION

Physiological outcomes

As the symptoms experienced by the patients with PEx occur on exercise, CPET has been hypothesised to be the best method of objectively quantifying changes in cardiopulmonary function that may result from surgery. CPET has evolved significantly over the period of this review. The initial articles published have used exercise testing in a less standardised manner than contemporary studies. More recent larger studies have used a more rigorous and detailed approach to CPET allowing for better comparison between studies. Of the 16 studies investigating CPET, 12 have shown significant improvements postoperatively. In children, the weight of evidence points to improvement. While earlier studies such as those by Wynn *et al* and Castellani *et al* failed to demonstrate significant improvement postoperative, the majority of more recent studies such as those by Maagaard, Das and Kelly studies have shown modest but significant gains both in maximum oxygen uptake and oxygen pulse. As well as demonstrating significant improvement in CPET indices following surgery, Maagaard also found that differences between PEx and healthy matched controls had resolved following Nuss bar removal. Most studies in children were of the Nuss procedure which reflects surgical trends in this age group. The Nuss bar remained in place at the end of the follow-up period in all but two of the studies, and further long-term follow-up is required to assess for further changes after removal.

Compared with paediatric patients, the cardiopulmonary impact of PEx surgical repair in adult patients is less well studied with only five studies published using

CPET. Among patients undergoing Ravitch procedure, significant improvements in exercise testing variables were evident from the early postoperative period.[22 26] With regard to the Nuss procedure, two small studies investigating patients 1 year after surgery with bar still in place showed contradictory results.[19 24] However, a larger more recent study by Jaroszewski *et al* has demonstrated significant and consistent improvements in all cardiopulmonary indices including an increase in maximum rate of oxygen consumption, oxygen pulse, oxygen consumption at anaerobic threshold and maximal ventilation ($p<0.001$ for all comparisons).[16] Even where results were nonsignificant, such as in Udholm *et al*, there was a tendency towards an increase in maximum oxygen uptake indicating that the adult patients might need a longer time to improve exercise capacity and lung function than paediatric population. VO_{2max} is also influenced by changes in weight and aerobic fitness which has the potential to confound these results. Only four of the studies attempted to control for this by objectively assessing for exercise habits before and after surgery.[12 19 20 22]

Of the 16 studies that assessment patients with CPET, only 4 did not demonstrate any benefit following surgery.[15 17 19 25] In all these studies bar Udholm *et al*, the mean VO_{2max} and O_2 pulse values were in the low normal range at baseline which may indicate less benefit for patients without clear exercise impairment preoperatively. All these studies were small however and may be underpowered to detect smaller differences in function postoperatively.

In terms of pulmonary function at rest, the data are often contradictory. The evidence is clear that surgery is not detrimental to resting pulmonary function long term, however, the benefit is less clear. Here, results have shown divergence between surgical techniques. Studies involving the Ravitch procedure were consistent in showing no change in pulmonary function after surgery.[14 22 33–36] With regard to the Nuss procedure, results have been more variable with different trends in PFTs depending on the time of follow-up and whether the bar has been removed. In the early postoperative period following Nuss procedure, pulmonary function was, in general, unchanged and may worsen. However, in all of these studies, values had normalised or improved on preoperative values after bar removal. Nine studies demonstrated improvement in pulmonary function after bar removal.[10 12 13 20 28 29 31 32 37] Improvements were driven by small but significant gains in FVC, FEV1 and total lung capacity (TLC); however, values were, in general, in the low normal range for the majority of patients. However, in the three studies to use control groups, all found preoperative pulmonary function to be significantly worse than aged matched healthy controls and that this difference improved or was even eliminated after bar removal.[10 12 13] This suggests that while not always falling in the pathological range, there is a limitation of pulmonary function in patients with PEx which can improve with surgery.

In general, the objective resting pulmonary function impairment of patients with PEx is mild and thus any improvement that can be expected from surgery would be modest. This makes measurement difficult, particularly in small studies which are, again, in general underpowered to detect small differences. This difficulty is confounded by the differing indications for surgery. Studies where the patients included had anatomically more severe PEx deformity with pre-existing PFT limitation such as that by Kelly *et al* and Tang *et al* showed an improvement postoperative which was not seen in patients operated on for purely cosmetic reasons such as in Aronson *et al* and Castellani *et al*. This may in part explain the divergent results between studies in this review.

As PEx is an anatomical issue with potential for cardiac compression, echocardiography has been widely studied in PEx patients. Generally, LV function does not appear to improve following surgery. Only one study demonstrated an increase in LV ejection fraction and in this study, ejection fraction is estimated only from a single plane, so changes may represent different positioning of the heart in the thoracic cavity as opposed to real change in LV function.[41] In terms of RV function, differences in results is evident between TTE and TOE procedures. TTE appears to be poor at measuring changes in RV in PEx patients and only two of the included studies showed improvement in RV function postsurgery. Distortion of normal cardiac geometry means that often optimal images may only be obtained through a subcostal window and complete evaluation of the heart is limited. In Laín *et al*, 20 patients showed evidence of compression during TOE, only 6 of which could be seen on preoperative TTE, which highlights the likelihood of underestimating cardiac compression which may in part reflect the divergent results between imaging techniques.[39] In contrast, TOE has shown more consistent improvement in cardiac indices. All four studies to use intraoperative TOE during repair have been consistent in demonstrating relief of cardiac compression. RV output was seen to increase by up 65% following repair. These findings suggest that, in PEx, cardiac filling and cardiac output might be limited by the result of compression by the displaced sternum on the right heart chambers, thereby limiting increase of stroke volume in response to exercise. They further demonstrate that this anatomical issue, much like the Haller index, can be remedied by surgery.

Since cardiac and respiratory systems are intimately related, it is difficult to disentangle the origin of the symptoms and pathophysiology of impairment, confusing efforts to show objective measurements of improvement after surgery. Pulmonary function at rest improves after Nuss procedure, however, modestly so and not to an extent that would explain improvement in symptoms seen after surgery or explain similar results in patients undergoing Ravitch procedure. Cardiac compression in PEx has been established and can be corrected with surgery but the extent to which this cardiac compression affects cardiopulmonary function and patients' symptoms has yet to be fully elucidated. Given the hypothesis that RV compression would produce a limitation in increasing stroke volume in response to exercise, CPET would be expected to reflect this with improvements in maximum O_2 uptake and O_2 pulse. This improvement has been demonstrated in children and more recently adults after both Ravitch and Nuss repair and offers a way of reproducibly and objectively measuring cardiopulmonary limitation in patients with PEx.

Psychological outcomes

The literature confirms the widely held view that psychological distress above the threshold for diagnosis of specific mental health disorder such as depressive illness or anxiety disorder is rare in PEx, or at least no more common than that of the general population. Of the five studies to specifically investigate mental health, only a single study identified patients falling above this threshold.

The improvement seen in QoL after pectus repair is manifest from the literature. All but one of the studies, that of Bahadir *et al*, showed significant improvement in QoL measures but this study had a number of key issues that limits its interpretation.[66] While it was prospectively carried out and used unoperated PEx patients as controls, it included a high proportion of patients with pectus carinatum and the groups were poorly matched. The unoperated group had higher preoperative body satisfaction so any changes are likely to reflect disease severity rather than the effect of surgery. The rest of the published studies are consistent in showing an improvement in QoL of patients with PEx using both generic and disease specific assessment tools. After repair, social activities were no longer affected by the patient's condition, and patients showed greater engagement in activities with family and peers.[50 53 56 59]

The majority of studies investigated QoL and psychological outcomes in children/adolescents undergoing Nuss repair, however, the Ravitch procedure and repair in adult patients have been shown to have comparable outcomes.[50 52 57] Kelly *et al* failed to show any correlation between disease severity and psychological and social issues which is supported by the wide variation in pectus severity seen in patients presenting with psychological complaints in the literature.[3] These findings highlight the importance of assessing body image and psychological distress in all patients with PEx and not just the anatomical severity. Patients with a minor deformity can suffer enormously and can feel restricted in their social life. Consequently, treatment could have a significant impact on the future of these patients.

While the literature indicates many improvements in psychological well-being following surgery, a threshold of psychological harm that justifies surgery has not yet been demonstrated, or the level below which surgery is of little benefit. This reflects the unrandomised nature of the included studies where all patients included (or their parents) perceived their distress or symptoms significant enough to electively consent to surgery.

Limitations

There are a number of limitations to this review. The review is broad in scope, attempting to cover all aspects of long-term outcomes of the two primary surgical methods of pectus repair, both in children and adults, looking at both cardiopulmonary and psychosocial outcomes. This is a strength, in that it allows a holistic approach to a topic where overly narrow approaches have failed to make definite conclusions. However, it also makes comparison between studies difficult given the wide and varying nature of outcomes studied. The studies included are heterogeneous in nature involving patients of different ages, undergoing different surgical methods with differing indications for surgery and degrees of disease severity. In particular, variability in disease severity has the potential for confounding. While the majority of studies relate to patients with anatomically moderate to severe disease there is considerable variability with mean Haller indices ranging from 3.9 to 9.6 and in 26 of the studies a different index is used or no index is recorded. The outcomes measured vary widely even within broad headings of physiological and psychological outcomes employed in this review. In the studies assessing psychological and QoL measures particularly, the terminology is opaque and technical reporting of psychometric questionnaires is difficult to interpret as a meaningful change to patient's lives.

The quality of data of the studies in the review is a limitation. The data quality is in general poor. No randomised control trials have been carried out on the surgical repair of PEx and such trials are unlikely to occur, given the reluctance of patients to leave a decision of 'surgery or not' to chance. This is particularly the case in PEx where patients (or parents) are often well researched and come seeking surgery. The majority of studies are small prospective studies which are in general underpowered to detect differences that may be modest in patients whose cardiopulmonary limitation falls around the lower range of normal. Larger retrospective cohort studies have been carried out such as that by Jaroszewski or Kelly *et al* but are at risk of confounding. The majority of studies are not well controlled. Only 7 of the 51 studies included in this review included a control group and in only in two studies, were patients with PEx not undergoing surgery used as the control group. Considering that the majority of patients in this review were children and adolescents, who are at a time of rapid physical and psychological change, controlling for changes in growth, fitness and self-perception is particularly important if meaningful conclusions are to be drawn. Particularly, when studying CPET, changes in aerobic conditioning can result in confounding and should be should be taken in context of exercise history. Only four of the included studies attempted to control for this.[12 19 20 22]

This issue with data quality and heterogeneity makes meta-analysis difficult and it was not attempted in this review. Four meta-analysis have been conducted to date on cardiopulmonary changes after PEx surgery, three assessing pulmonary function and one assessing cardiac function.[4 67–69] The results of these analyses were as varied as that of the studies from which they were derived. Jacobsen *et al* and Chen *et al* found modest improvement in pulmonary function after Nuss bar removal, a result which was not seen by Malek *et al*.[4 68 69] A second study of cardiovascular function by the same authors found significant improvements in pooled cardiovascular indices after repair, however, heterogeneity between studies makes interpretation difficult.[67] None assessed cardiopulmonary function during exercise and the scope of these reviews were much narrower than that of this present review.

CONCLUSION

The question this review has set out to answer is whether surgical repair improves cardiopulmonary and psychological outcomes in patients with PEx. While the existing literature is hampered by small, underpowered studies yielding often contradictory results, this review demonstrates that there are objective ways of measuring limitations in PEx and that they can be improved following surgery. While resting measures provide little yield in demonstrating this, exercise testing has been consistent in showing significant improvements in cardiopulmonary function following surgery, both for children and adults. This difference between rest versus exercise testing has led to contradictory results and what has been described as negative studies in some reports are in fact simply measuring the wrong outcomes. These demonstrated improvements in exercise testing indices have not yet been well correlated with patients symptoms which makes their clinical significance uncertain in terms of meaningfully outcomes for patients. What is clear, however, is the psychological impact that surgery can have. In almost all studies that measured it, symptom burden and QoL of patients with PEx improved significantly following surgery and the rate of satisfaction was high, generally greater than 90%. PEx is a disease established to have multifactorial effects on patient's lives and warrants a multifactorial assessment taking into account psychological and social elements as well as any physiological limitation that may be present.

Going forward, if this question is to be conclusively answered, it will require studies that, if not randomised, are at least well matched, using unoperated patients with PEx, prospectively carried out and controlling for changes with age in children and physical activity in adults. Minimally clinically important differences will need to be established to translate any improvement in physiological or psychological parameters into a guidance for clinicians in their decision-making whether to offer surgery or not.

The recent decision by the NHS to decommission surgery for PEx is likely to hamper efforts to conduct such a study and has far-reaching implications for patients with PEx. Historically, NHS commissioning has been based on need and QoL improvement however in its commissioning report, insufficient evidence is cited for improvement in physiological, psychological outcomes and QoL after surgery, a finding this review refutes. Perhaps, a practical step to answering this question and providing higher quality data would be to commence PEx in limited high volume centres in the UK with a dedicated outcome registry. An encouraging development in this regard is an interim commissioning pathway for PEx surgery been proposed by the NHS in April 2023. This would make repair available in a subset of patients with symptomatic and anatomically severe disease with clear evidence of cardiorespiratory impairment after multidisciplinary discussion.

A number of metanalyses have indicated the need for adequate surgical volume to reduce risk of complications from a surgery that many surgical trainees have little exposure to.[4 5] Thus, despite remaining unanswered for the best part of thirty years, the question over the surgical repair of PEx requires urgent answering or the chance to do so may be slipping away.

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REFERENCES

1 Fokin AA, Steuerwald NM, Ahrens WA, *et al*. Anatomical, histologic, and genetic characteristics of congenital chest wall deformities. *Semin Thorac Cardiovasc Surg* 2009;21:44–57.
2 Shamberger RC, Welch KJ, Castaneda AR, *et al*. Anterior chest wall deformities and congenital heart disease. *J Thorac Cardiovasc Surg* 1988;96:427–32.

3 Kelly RE, Cash TF, Shamberger RC, *et al*. Surgical repair of pectus excavatum markedly improves body image and perceived ability for physical activity: multicenter study. *Pediatrics* 2008;122:1218–22.
4 Johnson WR, Fedor D, Singhal S. Systematic review of surgical treatment techniques for adult and pediatric patients with pectus excavatum. *J Cardiothorac Surg* 2014;9:25.
5 Nasr A, Fecteau A, Wales PW. Comparison of the nuss and the ravitch procedure for pectus excavatum repair: a meta-analysis. *J Pediatr Surg* 2010;45:880–6.
6 Kelly RE, Shamberger RC, Mellins RB, *et al*. Prospective multicenter study of surgical correction of pectus excavatum: design, perioperative complications, pain, and baseline pulmonary function facilitated by Internet-based data collection. *J Am Coll Surg* 2007;205:205–16.
7 Jaroszewski DE. Physiologic implications of pectus excavatum. *J Thorac Cardiovasc Surg* 2017;153:218–9.
8 Shaalan AM, Kasb I, Elwakeel EE, *et al*. Outcome of surgical repair of pectus excavatum in adults. *J Cardiothorac Surg* 2017;12:72.
9 Page MJ, McKenzie JE, Bossuyt PM, *et al*. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *BMJ* 2021;372:n71.
10 Kelly RE, Mellins RB, Shamberger RC, *et al*. Multicenter study of pectus excavatum, final report: complications, static/exercise pulmonary function, and anatomic outcomes. *J Am Coll Surg* 2013;217:1080–9.
11 Aronson DC, Bosgraaf RP, Merz E-M, *et al*. Lung function after the minimal invasive pectus excavatum repair (Nuss procedure). *World J Surg* 2007;31:1518–22.
12 Maagaard M, Tang M, Nielsen HHM, *et al*. Normalized cardiopulmonary function following the Nuss procedure for Pectus Excavatum: 3-year follow-up. A prospective, controlled study. *Interact Cardiovasc Thorac Surg* 2012;15.
13 Tang M, Nielsen HHM, Lesbo M, *et al*. Improved cardiopulmonary exercise function after modified Nuss operation for pectus excavatum. *Eur J Cardiothorac Surg* 2012;41:1063–7.
14 Haller JA, Loughlin GM. Cardiorespiratory function is significantly improved following corrective surgery for severe pectus excavatum. *J Cardiovasc Surg (Torino)* 2000;41:125–30.
15 Wynn SR, Driscoll DJ, Ostrom NK, *et al*. Exercise cardiorespiratory function in adolescents with pectus excavatum. *J Thorac Cardiovasc Surg* 1990;99:41–7.
16 Jaroszewski DE, Farina JM, Gotway MB, *et al*. Cardiopulmonary outcomes after the nuss procedure in pectus excavatum. *J Am Heart Assoc* 2022;11:e022149.
17 Del Frari B, Blank C, Sigl S, *et al*. The questionable benefit of pectus excavatum repair on cardiopulmonary function: a prospective study. *Eur J Cardiothorac Surg* 2021;61:75–82.
18 Das BB, Recto MR, Yeh T. Improvement of cardiopulmonary function after minimally invasive surgical repair of pectus excavatum (Nuss procedure) in children. *Ann Pediatr Cardiol* 2019;12:77–82.
19 Udholm S, Maagaard M, Pilegaard H, *et al*. Cardiac function in adults following minimally invasive repair of pectus excavatum. *Interact Cardiovasc Thorac Surg* 2016;22:525–9.
20 O'Keefe J, Byrne R, Montgomery M, *et al*. Longer term effects of closed repair of pectus excavatum on cardiopulmonary status. *J Pediatr Surg* 2013;48:1049–54.
21 Maagaard M, Heiberg J. Improved cardiac function and exercise capacity following correction of pectus excavatum: a review of current literature. *Ann Cardiothorac Surg* 2016;5:485–92.
22 Neviere R, Montaigne D, Benhamed L, *et al*. Cardiopulmonary response following surgical repair of pectus excavatum in adult patients. *Eur J Cardiothorac Surg* 2011;40:e77–82.
23 Sigalet DL, Montgomery M, Harder J, *et al*. Long term cardiopulmonary effects of closed repair of pectus excavatum. *Pediatr Surg Int* 2007;23:493–7.
24 Cooper CB, Abrazado M, Saleh R, *et al*. Anatomical and physiological improvements following surgical correction of pectus excavatum: a prospective study. American Journal of Respiratory and Critical Care Medicine; 2010 181
25 Castellani C, Windhaber J, Schober PH, *et al*. Exercise performance testing in patients with pectus excavatum before and after nuss procedure. *Pediatr Surg Int* 2010;26:659–63.
26 Morshuis WJ, Folgering HT, Barentsz JO, *et al*. Exercise cardiorespiratory function before and one year after operation for pectus excavatum. *J Thorac Cardiovasc Surg* 1994;107:1403–9.
27 Cahill JL, Lees GM, Robertson HT. A summary of preoperative and postoperative cardiorespiratory performance in patients undergoing pectus excavatum and Carinatum repair. *J Pediatr Surg* 1984;19:430–3.
28 Sigalet DL, Montgomery M, Harder J. Cardiopulmonary effects of closed repair of pectus excavatum. *J Pediatr Surg* 2003;38:380–5.
29 Kuyama H, Uemura S, Yoshida A, *et al*. Pulmonary function in children with pectus excavatum and post-operative changes after nuss procedure. *Pediatr Surg Int* 2018;34:1099–103.
30 Kuru P, Dudakli A, Mursaloglu H, *et al*. How pulmonary function changes after pectus excavatum correction surgery. *Asian Cardiovasc Thorac Ann* 2015;23:945–9.
31 Kubiak R, Habelt S, Hammer J, *et al*. Pulmonary function following completion of minimally invasive repair for pectus excavatum (MIRPE). *Eur J Pediatr Surg* 2007;17:255–60.
32 Lawson ML, Mellins RB, Tabangin M, *et al*. Impact of pectus excavatum on pulmonary function before and after repair with the nuss procedure. *J Pediatr Surg* 2005;40:174–80.
33 Jiang X, Hu T, Liu W, *et al*. Pulmonary function changes following surgical correction for pectus excavatum. *Chin Med J (Engl)* 2000;113:206–9.
34 Kowalewski J, Barcikowski S, Brocki M. Cardiorespiratory function before and after operation for pectus excavatum: medium-term results. *Eur J Cardiothorac Surg* 1998;13:275–9.
35 Morshuis W, Folgering H, Barentsz J, *et al*. Pulmonary function before surgery for pectus excavatum and at long-term follow-up. *Chest* 1994;105:1646–52.
36 Kaguraoka H. A study of respiratory function, before and after surgery, in patients with funnel chest. *Journal of Tokyo Women's Medical College* 1990;60:494–504.
37 Szydlik S, Jankowska-Szydlik J, Zwaruń D, *et al*. An effect of nuss procedure on lung function among patients with pectus excavatum. *Pol Przegl Chir* 2013;85:1–5.
38 Gürkan U, Aydemir B, Akgöz H, *et al*. Echocardiographic assessment of right ventricular functions before and after surgery for Pectus Excavatum. *Turk Kardiyol Dern Ars* 2012;40.
39 Laín A, Giralt G, Giné C, *et al*. Transesophageal echocardiography during pectus excavatum correction in children: what happens to the heart? *J Pediatr Surg* 2021;56:988–94.
40 Chao CJ, Jaroszewski D, Kumar P, *et al*. Results of Pectus Excavatum repair surgery-just cosmetic or of structural and functional benefit? *J Am Soc Echocardiogr* 2015;28:B116.
41 Krueger T, Chassot P-G, Christodoulou M, *et al*. Cardiac function assessed by transesophageal echocardiography during pectus excavatum repair. *Ann Thorac Surg* 2010;89:240–3.
42 Coln E, Carrasco J, Coln D. Demonstrating relief of cardiac compression with the nuss minimally invasive repair for pectus excavatum. *J Pediatr Surg* 2006;41:683–6.
43 Neviere R, Wurtz A. Longer term effects of closed repair of pectus excavatum on cardiopulmonary status. *J Pediatr Surg* 2013;48:1988–9.
44 Chao C-J, Jaroszewski DE, Kumar PN, *et al*. Surgical repair of pectus excavatum relieves right heart chamber compression and improves cardiac output in adult patients--an intraoperative Transesophageal echocardiographic study. *Am J Surg* 2015;210:1118–24.
45 Töpper A, Polleichtner S, Zagrosek A, *et al*. Impact of surgical correction of pectus excavatum on cardiac function: insights on the right ventricle. A cardiovascular magnetic resonance study†. *Interact Cardiovasc Thorac Surg* 2016;22:38–46.
46 Schaarschmidt K, Polleichtner S, Zagrosek A, *et al*. Quantifying hemodynamic effects of pectus excavatum and nuss repair on right and left ventricular function by high speed "cardiovascular magnetic resonance cine-Ssfp-imaging," early results of the prospective berlin-buch nuss-Cmr study. *J Laparoendosc Adv Surg Tech* 2011;21:A22–3.
47 Peterson RJ, Young WG, Godwin JD, *et al*. Noninvasive assessment of exercise cardiac function before and after pectus excavatum repair. *J Thorac Cardiovasc Surg* 1985;90:251–60.
48 Blickman JG, Rosen PR, Welch KJ, *et al*. Pectus excavatum in children: pulmonary scintigraphy before and after corrective surgery. *Radiology* 1985;156:781–2.
49 Lawson ML, Cash TF, Akers R, *et al*. A pilot study of the impact of surgical repair on disease-specific quality of life among patients with pectus excavatum. *J Pediatr Surg* 2003;38:916–8.
50 Krasopoulos G, Dusmet M, Ladas G, *et al*. Nuss procedure improves the quality of life in young male adults with Pectus Excavatum deformity. *Eur J Cardiothorac Surg* 2006;29:1–5.
51 Metzelder ML, Kuebler JF, Leonhardt J, *et al*. Self and parental assessment after minimally invasive repair of pectus excavatum: lasting satisfaction after bar removal. *Ann Thorac Surg* 2007;83:1844–9.
52 Lam MWC, Klassen AF, Montgomery CJ, *et al*. Quality-of-life outcomes after surgical correction of pectus excavatum: a comparison of the ravitch and nuss procedures. *J Pediatr Surg* 2008;43:819–25.

53 Jacobsen EB, Thastum M, Jeppesen JH, *et al*. Health-related quality of life in children and adolescents undergoing surgery for pectus excavatum. *Eur J Pediatr Surg* 2010;20:85–91.
54 Bostanci K, Ozalper H, Karadeniz PG, *et al*. Quality of life outcomes after minimally-invasive repair of pectus excavatum. *Interact Cardiovasc Thorac Surg* 2011;13:S15.
55 Hadolt B, Wallisch A, Egger JW, *et al*. Body-image, self-concept and mental exposure in patients with pectus excavatum. *Pediatr Surg Int* 2011;27:665–70.
56 Kim HK, Shim JH, Choi KS, *et al*. The quality of life after bar removal in patients after the nuss procedure for pectus excavatum. *World J Surg* 2011;35:1656–61.
57 Kuru P, Bostanci K, Ermerak NO, *et al*. Quality of life improves after minimally invasive repair of pectus excavatum. *Asian Cardiovasc Thorac Ann* 2015;23:302–7.
58 Sacco Casamassima MG, Gause C, Goldstein SD, *et al*. Patient satisfaction after minimally invasive repair of pectus excavatum in adults: long-term results of nuss procedure in adults. *The Annals of Thoracic Surgery* 2016;101:1338–45.
59 Lomholt JJ, Jacobsen EB, Thastum M, *et al*. A prospective study on quality of life in youths after pectus excavatum correction. *Ann Cardiothorac Surg* 2016;5:456–65.
60 Luo L, Xu B, Wang X, *et al*. Intervention of the nuss procedure on the mental health of pectus excavatum patients. *Ann Thorac Cardiovasc Surg* 2017;23:175–80.
61 Wachter T, Del Frari B, Edlinger M, *et al*. Aesthetic outcomes after surgical repair of pectus excavatum in females: differences between patients and professional evaluators. *Arch Plast Surg* 2020;47:126–34.
62 Zuidema WP, Oosterhuis JWA, Zijp GW, *et al*. Sports activity in adolescents in the Netherlands with a pectus excavatum; the impact of surgery. *J Pediatr Surg* 2019;54:1671–4.
63 Norlander L, Sundqvist AS, Anderzén-Carlsson A, *et al*. Health-related quality of life after nuss procedure for pectus excavatum: a cross-sectional study. *Interact Cardiovasc Thorac Surg* 2022;35:ivac031.
64 Bahadir AT, Kuru P, Afacan C, *et al*. Validity and reliability of the Turkish version of the nuss questionnaire modified for adults. *Korean J Thorac Cardiovasc Surg* 2015;48:112–9.
65 Zuidema WP, Oosterhuis JW, van der Heide SM, *et al*. Correlation of preoperative state anxiety and pain six weeks after surgical correction of pectus excavatum. *Anesth Pain Med* 2020;10:e98969.
66 Bahadir AT, Kuru Bektaşoğlu P, Çakiroğlu Eser A, *et al*. Psychosocial functioning in pediatric patients with pectus excavatum and pectus carinatum. *Turk J Med Sci* 2017;47:771–7.
67 Malek MH, Berger DE, Housh TJ, *et al*. Cardiovascular function following surgical repair of pectus excavatum: a metaanalysis. *Chest* 2006;130:506–16.
68 Malek MH, Berger DE, Marelich WD, *et al*. Pulmonary function following surgical repair of pectus excavatum: a meta-analysis. *Eur J Cardiothorac Surg* 2006;30:637–43.
69 Chen Z, Amos EB, Luo H, *et al*. Comparative pulmonary functional recovery after nuss and ravitch procedures for pectus excavatum repair: a meta-analysis. *J Cardiothorac Surg* 2012;7:101.

Appendix

Appendix 1. Search Strategy

PubMed:

SET A: Pectus excavatum
"Funnel Chest"[Mesh] OR "Funnel Chest"[Text Word] OR "Pectus Excavatum"[Text Word] OR "Funnel Breast"[Text Word] OR "Cobbler's chest"[Text Word] OR "Sunken chest"[Text Word]

n = 3,391

SET B: Surgery
“Thoracic Surgery” [Mesh] OR "Thoracic Surgical Procedures" [Mesh] OR (Repair[Text Word] OR "Pectus Surgery"[Text Word] OR Nuss[Text Word] OR Ravitch[Text Word] OR "Minimally Invasive"[Text Word] OR "Open Surgery"[Text Word] OR Surgery[Text Word] OR "Surgical Intervention"[Text Word])

n = 3,338,415

A + B = **2267**

Embase:

Set A

1. 'funnel chest'/exp OR 'funnel chest'	4682
2. 'pectus excavatum':ti,ab,kw	3122
3. funnel AND breast:ti,ab,kw	431
4. #1 OR #2 OR #3	5309

Set B

5. ‘Thoracic surgery'/exp OR 'thoracic surgery' OR (thoracic AND ('surgery'/exp OR surgery))	931544
6. Thorax surgery	43,581
7. 'pectus surgery':ti,ab,kw	21
8. Nuss:ti,ab,kw	1073
9. Ravitch:ti,ab,kw	367
10. 'surgery'/exp OR surgery:ti,ab,kw	6,072,446
11. 'surgical intervention':ti,ab,kw	87087
12. 'minimally invasive procedure' OR 'minimally invasive':ti,ab,kw	128733
13. 'open repair':ti,ab,kw	7477
14. Repair:ti,ab,kw	481477
15. #5 OR #6 OR #7 OR #8 OR #9 OR #10 OR #11 OR #12 OR #13 OR #14	6,420,331

Set A + B

16. #4 AND #15	**3,196**

Cochrane

"Funnel Chest"[Mesh] OR "Funnel Chest"[Text Word] OR "Pectus Excavatum"[Text Word]

n = 1

Supplementary Table 1: Cardiopulmonary Outcomes Following Pectus Repair

Year	Study	Design	Patient Demographics	Mean Haller Index	Follow up post-surgery (months)	Bar/ No bar	Cardiopulmonary outcomes	Results
2022	Jaroszewski	Retrospective cohort study Uncontrolled Single centre	n = 130 Age 32.4 ±10 Nuss	4.6 ±2.2	36	Bar	CPET	Significant increase in VO_{2max}, 26.1±6.8 to 29.1±8.5 ml/kg/min (70.8±17.0 to 82.5±22.0 % predicted) and significant increase in O_2 pulse 11.8±3.4 to 13.1±3.8 ml/beat (83.0±17.4 to 92.2±24.2 % predicted).
							TOE (n =36)	Significant increase in RV function: RVOT 14.2±3.3cm to 16.3±3.4cm (p< 0.005) and RV SV 41.1±13.2ml to 54.6±15.6 (p < 0.001).
2021	Lain	Prospective cohort study Uncontrolled Single centre	n = 20 Age 13.5± 2.9 Nuss	6.3 ±2.63	Intraoperative	Bar	TOE	Significantly increase in diameter of right ventricle was 5.78 +/- 3.5mm (p < 0.05), right atrium 6.64 +/- 5mm (p < 0.05) and tricuspid annulus 6.02+/- 3.29mm (p < 0.05).
2021	Del Frari	Prospective cohort study Uncontrolled Single centre	n = 19 Age 13.9-19.6 Nuss	4.2 ± 1.0	9	Bar	PFT	No significant change in variables.
							TTE	No significant change in variables: RVEDV 32.0 ± 2.57mm and LVEDV 45.0 ± 4.67mm baseline w/ no change post-op.
							CPET	No significant change in variables: VO_{2max} 41.0 ± 6.2 to 40.4 ± 4.0 ml/kg/min.
2019	Das	Retrospective cohort study Uncontrolled Single centre	n = 24 Age 12.9±3.6 Nuss	4.3±0.9	24	Bar	CPET	Significant improvement in VO_{2max} by 40.6%: 32 ± 13 to 45 ± 10 ml/kg/min (p = 0.0001) Significant improvement in oxygen pulse by 44.4%: 9 ± 4–13 ± 5 ml/beat (p = 0.03).
2018	Kuyama	Retrospective cohort study Uncontrolled Single centre	n = 43 Paediatric Nuss	4.3 ± 1.3	48	No bar	PFT	FVC fell post bar insertion but significantly increased 1 year post bar removal.

							CPET	No significant change in variables: VO_{2max} 30.4 ± 1.9 to 33.3 ± 1.6 ml/kg/min(p = 0.0940).
2016	Udholm	Prospective cohort study Uncontrolled Single centre	n = 15 Age 32 Nuss	NR	12	Bar	PFT	No significant change in variables: FVC 90±9 to 88±7% (p = 0.2731), FEV1 89±10 to 90±9 %/predicted (p = 0.7896).
2015	Kuru	Prospective cohort study Uncontrolled Single centre	n = 80 Age 16.91 ± 4.37 Nuss	4.07 ± 1.4	6	Bar	PFT	No significant change in FEV1. Significant decrease in FVC decreased from 83.21% ± 16.97% to 76.52% ± 20.98% (p = 0.01).
2016	Toepper	Prospective cohort study Uncontrolled Single centre	n = 38 Age 21±8.3 Nuss	9.64± 22	12	Bar	cMRI	Significant decrease in heart rate (p < 0.0001) Significant increase in RVEF (p = 0.0004), RVSV (*P* = 0.0167), LVEF (p = 0.0165), and LVSV (p = 0.0036). No significant change in RVEDV (p = 0.7590), RVESV (p = 0.0718), LVEDV (p = 0.0648), and in LVESV (p = 0.8135).
2015	Chao et	Retrospective cohort study Uncontrolled Single centre	n = 168 Age 33 (18-71) Nuss	5.7 ± 3.1	Intraoperative	Bar	TOE	Significant improvement in RA (15.1%), tricuspid annulus end systolic (10.9%), right ventricular outflow tract end diastolic (6.1%) and end systolic dimension (6.1%) size after surgery (all P < 0.0001). Significant improvement in right ventricular cardiac output by 38% (P < 0.0001).
2014	Gurkan	Retrospective cohort study Uncontrolled Single centre	n = 16 Age 20.5 ± 5.6 Nuss	NR	1	Bar	TTE	Significant increase in RV end-diastolic diameter, tricuspid annular plane systolic excursion (TAPSE), pulsed tissue Doppler systolic velocity (S'). Significant improvement in myocardial performance indexes of both the right and the left ventricles.
2013	Szydlik	Retrospective cohort study Uncontrolled Single centre	n = 44 Age 16 (10-32) Nuss	NR	40	No Bar	PFT	Significant improvement in FVC, 75 to 89% (P = 0.00016) and FEV1, 85% to 95% (p =00125).

2013	O'Keefe	Prospective cohort study Uncontrolled Single centre	n = 67 Age 13.9 ± 2.3 Nuss	4.4 ± 1.3	42	No bar	PFTs CPET TTE Subjective exercise tolerance	Significant improvements in FEV1, 81±17 to 89. ±20.5 % predicted (p < 0.001) and FVC, 91.2±18.6 to 98.9 ± 22 % predicted (p < 0.01). Significant improvements in O_2 pulse 75.8 ± 14.4 vs 80.5 ± 18.3* % predicted (P < 0.01) but no significant change in VO_{2Max}, 33.2±7.5 to 34.2±7.5ml/kg/min (p = 0.09). No significant change in variables. Subjective ability to exercise (3.3± 0.7 vs 4.3 ± 0.6, scale 1–5) increased significantly.
2013	Maagard	Prospective cohort study **Controlled (age matched healthy controls)** Single centre	n = 68 (42paitents, 24 controls) Age 15.5 ± 1.7 Nuss	4.9 ±1.4	36	No bar	PFT TTE CPET	Low normal function in patients pre-operatively w/ significant improvement in FEV1 1-year (p = 0.001) and 3-year follow-ups (p = 0.0001), patients improved FEV1 compared with baseline and were no longer different from controls. No significant difference between patients and controls or pre and post-surgery. Before correction, CImax was lower in patients compared with controls during exercise, 6.6±1.2 vs 8.0±1.7 mL/min/m2. Increased both from 1-year to 3-year follow-up (p = 0.001) and from baseline to 3-year follow-up (p = 0.0001). After bar removal at the 3-year follow-up, CImax had normalized and no significant difference was seen between patients and controls, 8.1±1.2 and 8.3±1.6 (p = 0.572).
2013	Kelly	Prospective cohort study **Controlled (age matched healthy controls) Multicentre**	n= 182 / n = 20 13.56 ± 3 Nuss/Ravitch	4.4	12	Bar	PFT CPET	Significant increase in FVC from 88% to 93%, FEV1 from 87% to 90%, and TLC from 94% to 100% of predicted (p < 0.001 for each). VO_{2max} increased by 10.1%, 3.18±0.3 to 3.50±0.3 L/min (p = 0.015) and O_2 pulse by 19%, 13.58±3 to 16.16±4.9 (p = 0.007).

2012	Tang	Prospective cohort study **Controlled (age matched healthy controls)** Single centre	n = 75 (49 patients, 26 controls) Age 15.5 ± 1.7 Nuss	5.3 ± 2.3	12	Bar	PFT TTE CPET	FEV1 was low normal (87%) w/ significant difference between patients and controls (94%) which increased significantly to 91% 1 year post op (P < 0.01) however still remained lower than controls. No significant change in variables. No significant change in VO_{2max}, 26±6 to 28±6 ml/kg/min, Maximum cardiac index was significantly improved at 1 year postoperatively, 6.6± 1.1 to 7.2± 1 L/min/m2 (P = 0.0054) but was still significantly lower than the control group (P = 0.0008).
2012	Schaarschmidt	Prospective cohort study Uncontrolled Single centre	n = 51 Age 21.1±8.6 Nuss	8.3±3.6	12	Bar	CMR	Right and left ventricular ejection fraction and right ventricular stroke volume (SV) are significantly increased 2 weeks (p < 0.001) and 3 month (p < 0.001) after Nuss (similar trend in the few patients 1 year post).
2011	Neviere	Prospective cohort study Uncontrolled Single centre	n = 70 Age 27 ± 11 Ravitch	4.7 ± 1.4	12	Ravitch	PFT TTE CPET	No significant change in variables. No significant change in variables. Significant increase in VO_{2max} 34.9±7 to 37.6±7 (77 ±2% to 87±2 % predicted) (P < 0.0005) and O_2 pulse 13.2±3 to 14.8± 3 (P< 0.003).
2010	Krueger	Prospective cohort study Uncontrolled Single centre	n = 17 Age 28 (17–54) Ravitch	NR	Intraoperative	Ravitch	TOE	Significant increase in RVED diameter, area, volume; 2.4 ± 0.8 cm versus 3.0 ± 0.9 cm, (p < 0.001); 12.5 ± 5.2 cm2 versus 18.4 ± 7.5 cm2, (p < 0.001); and 21.7 ± 11.7 mL versus 40.8 ± 23 mL, (p < 0.001) and LV function 58.4% ± 15% versus 66.2% ± 6%, (p < 0.001).
2010	Sigalet	Prospective cohort study Uncontrolled Single centre	n = 26 Age 13.2 ± 2.1 Nuss	4.5 ± 1.3	40	No bar	PFT CPET	Significant improvement in FEV1 78 ± 16 to 84 ± 18 % predicted (p < 0.05) and TLC 95 ± 16 to 99 ± 14 % predicted (p < 0.05), no significant change in FVC 89.5 ± 18 to 92.4 ± 20 % predicted. Significant improvement in VO_{2max} 71 ± 11 to 77 ± 11 % predicted, (p < 0.05) and O_2 Pulse 77.1 ± 9.5 to 82.5 ± 9.2 % predicted (p < 0.05).

2010	Cooper	Prospective cohort study Uncontrolled Single centre	n = 28 Age 24 ± 14 Nuss	8.89±5	9	Bar	PFT cMRI CPET	No significant change in TLC. RVSV increased by 11 ml (p = 0.005). VO_{2max} 2.37 to 2.6 L/min (p = 0.002).
2010	Castellani	Prospective cohort study Uncontrolled Single centre	n = 59 Age 15.7 ± 4.5 Nuss	Different index	3.5	No bar	PFT CPET	Transient FVC decreased from 91% of normal value to 79%, but again increased to 88% after implant removal (no significant difference long term.) No significant change in VO_{2max} 42.6±6to 42.2±7.2 ml/kg/min.
2007	Kubiak	Retrospective cohort study Uncontrolled Single centre	n = 15 Age 15.9 Nuss	6 ± 2.1	40	No bar	PFT	Significant improvement in FVC 63.7 to81.3 % peracted (p = 0.03), FEV1 62.9 to 84.7% %predicted (p =0.03) and the RV/TLC ratio, 173.7 to 118.9 (p < 0.001).
2007	Aronson	Prospective cohort study Uncontrolled **Multicentre**	n = 145 Age 14.9 ± 6 Nuss	NR	36	No bar	PFT	No significant change in PFTS from pre-op values after bar removal. PFTs were normal pre-op and majority of patients were operated on for cosmetic reasons.
2006	Coln	Retrospective cohort study Uncontrolled Single centre	n = 123 Age 13 (5-18) Nuss	4.3±1.6	24	Bar	TTE (during exercise)	Cardiac compression in 117 (95%) and mitral valve abnormality was present in 54 (44%) on TTE during exercise Significant reduction in cardiac compression, normal post-op echo in 100 patients (93%).
2005	Lawson	Retrospective cohort study Uncontrolled Single centre	n = 45 Age 11.4 Nuss	4.6 ± 2	48	No Bar	PFT	Preoperatively, FVC and FEV1 medians were lower than the normal by 13%, whereas the FEF25-75 median was lower than normal by 20% (all P < 0.01). Small but significant improvement: 6% improvement in FVC, 9% improvement in FEV1, and a 15% improvement in FEF25-75.
2000	Jiang	Prospective cohort study Uncontrolled Single centre	n = 27 Age: 8.6 (3-16) Ravitch	NR	24	Ravitch	PFT	No significant change in variables.
2000	Haller	Prospective cohort study		NR	6	Ravitch	PFT	Lower FVC in patients versus controls but no significant change post repair.

		Controlled (age matched healthy controls) Single centre	46 (36 patients and 10 controls) Age 16 ± 3 Ravitch				Exercise testing	Significant increases in duration ($P < 0.02$), speed ($P < 0.02$), and grade of exercise ($P < 0.05$) and in O_2 pulse ($P < 0.005$).
1998	Kowaleski	Retrospective cohort study Uncontrolled Single centre	34 (22 moderate, 12 severe) Age 13 (4-35) Ravitch	Different index used	5	Ravitch	PFT TTE	Significant improvement in FEV1 only in those w/ severe ventilatory impairment pre-operatively. No improvement in LV function but significant improvement RV diastolic volume and SV index following surgery.
1994	Morshuis	Prospective cohort study Uncontrolled Single centre	n = 35 Age 17.9 ±5.6 Ravitch	NR	12	Ravitch	PFT Exercise testing	TLC 86.0% +/- 14.4%; (p = 0.0001) and FVC 79.7% +/- 16.2; (p = 0.0001) were significantly smaller than predicted and further decreased after operation -9.2% +/- 9.2%; (p = 0.0001) and -6.6% +/- 10.7% (p = 0.0012). Significant increase in VO_{2max} (p =0.0002) and O_2 pulse (p = 0.0333).
1992	Kaguraoka	Retrospective cohort study Uncontrolled Single centre	n = 138 Age 19.6 ± 7.6 Ravitch	Different index	42	Ravitch	PFT	Significant decrease in FVC by10 % early post-op with no significant change in variables from baseline long term.
1990	Wynn	Prospective cohort study **Controlled (PEx not undergoing surgery)** Single centre	12 (8 patients, 4 PEx controls) Age 13.6 Ravitch	Different index	12	Ravitch	Exercise testing	No significant changes pre or post operation: VO_{2max} 36.1 ± 4.4 to 38.1 ± 8.1 ml/kg/min in study group and 41.2 ± 7.3 to 43.0 ± 6.9ml/kg/min in control group.
1985	Peterson	Prospective cohort study Uncontrolled Single centre	n = 13 Age 13 ± 4 Ravitch	Different index	6	Ravitch	First pass radionucleotide cardiography	No changes in LVEF or cardiac index at rest or exercise, significant increase in RV end diastolic volume.

1985	Blickman	Prospective cohort study Uncontrolled Single centre	n = 17 Age 12.3 (4-21) Ravitch	NR	6	Ravitch	Perfusion and ventilation scintigraphy	Ventilation-perfusion ratios were abnormal in 10 of 17 patients, normalizing postoperatively in 60%.
1984	Cahill	Prospective cohort study Uncontrolled Single centre	n = 14 Age 11± 3 Ravitch	NR	6	Ravitch	Exercise testing	Significant improvement in maximal voluntary ventilation ($p < 0.001$), improvement in exercise performance quantified by the total exercise time and the maximal oxygen consumption ($p < 0.01$).

NR indicates not recorded; CPET, Cardiopulmonary exercise testing; PFT, Pulmonary function testing; TTE, Transthoracic echocardiography; TOE, Transoesophageal echocardiography, VO_{2max}; Maximum oxygen consumption; FVC; Forced vital capacity, FEV1; Forced expiratory volume in one second RV; Right ventricle; LV; Left ventricle

Supplementary Table 2: Psychological & Quality of Life Outcomes Following Pectus Repair

Year	Study	Design	Patient Demographics	Mean Haller Index	Follow up (months)	Outcomes (Assessment tool)	Results
2022	Norlander	Cross-sectional study Multicentre	n = 236	NR	NR	Specific HRQoL (modified Nuss Questionnaire)	Comparison to pre-op/unoperated patients, found significantly better HRQoL in patients who had surgery at a younger age.
2019	Zuidema	Prospective cohort study Uncontrolled Single centre	n = 127 Age 16.2± 2 Nuss	3.9± 1.4	12	Sports activity (CHQ, SF-36 and PEEQ)	No significant change in participation in sport (67% vs 65%) one year after surgery. Significant decrease in post-operative physical complaints during sports activity (p < 0.001).
2019	Wachter	Prospective cohort study Uncontrolled Single centre	n = 18 Age 20± 2 Nuss	NR	58	Aesthetic and psychological outcome (local questionnaire)	Patients rated their disease as more severe than medical professionals pre-operatively. Significant improvement in self-perception and psychological wellbeing post bar removal.
2017	Luo	Prospective cohort study Uncontrolled Single centre	n = 266 Age 19 ± 4 Nuss	4.1 ± 1	12	Psychiatric and psychological symptoms (SCL-90) Depressive illness (SDS)	Significant reduction in mental health problems from 161 (60.53%) to 79 (29.70%) (P<0.001). Significant reduction in depressive illness from 153 (57.52%) to 76 (28.57%) (p <0.001).
2017	Bahadir	Prospective study **Controlled** Single centre	n = 63 (32 operated / 31 controls) Age NR NR	NR	6	Psychiatrics and psychological symptoms (CDI, SDQ)	No significant difference between operated and un-operated patients.
2016	Lomholt	Prospective cohort study Controlled (age matched healthy controls) Single centre	n = 107 Age 15±2 Nuss	NR	12	Generic HRQoL (CHQ)	Comparable overall HRQoL pre-operatively to that of health controls w/ exception of perceived physical functioning (P< 0.0001). Significant improvement improved emotional wellbeing and self-esteem, as well as an increase in physical and social activities from pre- to post-surgery (P<0.001).
2016			n = 39	4.3 ± 1.3	40	Patient satisfaction	Patient satisfaction of 82%.

	Sacco Casamassima	Retrospective case series Single centre	Age 30 (21 to 55.) Nuss			Specific HRQoL (mNuss Questionnaire)	90% self-reported improvement in general health, exercise tolerance and social interaction.
2016	Kuru	Prospective cohort study Uncontrolled Single centre	n = 88 Age 18 ± 4 Nuss	4.04 ± 1.34	6	Disease specific HRQoL (mNuss Questionnaire) Psychosocial functioning	Median scores of 31 (31-35) to 43 (43-46) (P<0.001). Psychosocial functioning: median, interquartile range): pre-surgery 22.5 (19 to 25), 6 months post-surgery 33 (30-35)) (P<0.001).
2011	Kim	Prospective cohort study Uncontrolled Single centre	n = 39 Age 7 ± 3 Nuss	NR	42	Patient satisfactions and social wellbeing pre-operatively, post-operatively and post Nuss bare removal (non-validated local questionnaire)	Significant increase in satisfaction and social participation post operatively sustained post bar removal.
2011	Hadolt	Prospective cohort study Uncontrolled Single centre	n = 17 Age 19±2.5 Nuss	NR	48	Operative expectations (OPE Questionnaire) Body Image (FKKS) Psychiatric and psychological symptoms (SCL-90)	Expectations were meant in 82% of cases. Significant improvement. Pre-operatively patient scores were within the normal range however there were significant improvements in depression (P<0.05), Global severity index (P<0.03) and positive symptom distress burden (P<0.05) within this range.
2011	Bostanci	Prospective cohort study Uncontrolled Single centre	n = 140 Age 16 (6-35) Nuss	NR	30	Disease specific HRQoL (mNuss Questionnaire) Patient satisfaction	Statistically significant improvement (P<0.0001) on the overall quality of life, both in psychosocial (P<0.0001) and physical (P<0.0001) components. Patient satisfaction was 95%.

2010	Jacobson	Prospective cohort study Controlled Single centre	n = 119 271 controls Age (8-20)	NR	12	Generic HRQoL (CHQ)	Significantly better scores in family participation and mental health versus healthy controls.
2008	Kelly	Prospective cohort study Uncontrolled Multicentre	n = 326 Age (8-21) 291 parents Nuss (283) / Ravitch (43)	4.4± 1	12	Disease specific HRQoL (Nuss Questionnaire)	No correlation between degree of severity (PSI) and physical or psychosocial difficulties. Patients: Body image, physical difficulties significantly improved postoperatively. Parents: Emotional, social and physical difficulties significantly improved postoperatively.
2008	Lam	Retrospective cohort study Uncontrolled Single centre	n = 43 Age 15 ± 2 Ravitch 22 / Nuss 19	4.0 ± 0.9	15	Generic HRQoL (CHQ) Disease specific HRQoL (Nuss Questionnaire)	No significant difference in HRQL outcomes between Nuss and Ravitch procedures. Post-operatively reported lower CHQ scores relative to normative population.
2007	Metzelder	Prospective cohort study Uncontrolled Single centre	n = 44 Age 13.5 (6-20) Nuss	NR	54	Disease specific HRQoL (Nuss Questionnaire) Patient satisfaction	Significant improvement of psychosocial and physical well-being after bar implantation, which persisted up to 4 years after bar removal (p < 0.001). Patient satisfaction was 94%.
2006	Krasopoulos	Prospective cohort study Uncontrolled Single centre	n = 20 Age 18 (14-27) Nuss	NR	5	Disease specific HRQoL (mNuss Questionnaire)	Statistically significant improvement (p=0.001) in social function, self-esteem and a high level of satisfaction following the Nuss procedure. Statistically significant improvement only for the degree of dyspnoea (question 15, p = 0.005).

2003	Lawson	Prospective cohort study Uncontrolled Single centre	n = 19 Age (8-18) Nuss	NR	12	Disease specific HRQoL (Nuss Questionnaire)	Significant improvements in body image, exercise intolerance, shortness of breath, and tiredness and frequency of the child being frustrated, sad, self-conscious, and isolated by both patients and parents.

NR indicates Not recorded; QoL; Quality of Life; HRQoL; Health related quality of life; CHQ; Children's health questionnaire; SF-36; Short Form-36; PEEQ; Pectus Excavatum Evaluation Questionnaire; SDS, Self-rating depression scale; SDQ, Strengths and difficulties questionnaire; SCL-90, Symptom checklist-90; FKKS, Frankfurt body concept scale