Evidence Review:

Surgical correction for pectus deformity (all ages)

NHS England

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Contents

1. Introduction

Pectus abnormalities cover a range of deformities affecting the anterior thorax, specifically the sternum and adjacent rib cartilages. The incidence is less than 10 per 1,000 population, with the vast majority of patients being affected to only a very minor degree.

Pectus abnormalities arise due to an unevenness of the growth of the chest wall and are therefore commonest in teenagers and young adults, but can also affect children. It is more common in males. There are two main types of deformity – pectus excavatum and pectus carinatum, the former being the commoner of the two. Typically they are isolated deformities but they are sometimes associated with other musculoskeletal or connective tissue abnormalities such as scoliosis, Poland's syndrome and Marfan's syndrome. They can be familial.

Most pectus deformities become apparent in the first decade of life but are often not noticed until the adolescent growth spurt. If not corrected, the deformity is permanent.

There are currently a number of surgical and non surgical techniques available and individuals with a pectus deformity may be referred to a thoracic surgical clinic for advice. Based on NHS Secondary Users Services data, it is estimated that approximately 380 pectus surgery operations are performed by the NHS in England each year.

This policy considers the evidence for two surgical procedures used to correct pectus deformity - Nuss (minimally invasive repair of pectus excavatum – MIRPE) and Ravitch. The Nuss procedure is generally only applicable in pectus excavatum whilst the Ravitch procedure can be used for both pectus excavatum and pectus carinatum.

2. Summary of results

The evidence review of surgical correction of pectus excavatum using the Nuss procedure (minimally invasive repair of pectus excavatum) or the Ravitch/modified Ravitch procedure (open thoracic surgery) was undertaken with a view to answer the following research questions:

- Is there evidence that surgical correction improves cardiorespiratory reserve and functionality for the patient?
- Is there evidence that surgeon volume impacts on the outcomes of surgery (infection and revision rates)?
- What is the evidence in terms of quality, safety and adverse events associated with surgical correction?
- Is there evidence relating to eligibility and thresholds for surgery?

In summary, the current body of clinical evidence is largely limited to case series and reports. As such, the systematic reviews and meta -analysis of these observational studies are at risk of significant bias and confounding. Most studies do not attempt to address statistical heterogeneity between studies or take into account surgical skill variations amongst individual surgeons, between centres and over time. The absence of a standardised measure/scale to weigh clinical benefits (physical, psychological and quality of life) against the significant morbidity caused by the procedures presents a challenge to any conclusion regarding benefits of the intervention.

Cardiorespiratory reserve, functional and physical outcomes:

Johnson et al, 2014 found no linkage between ages of operative treatment with outcomes. There was no clear difference in outcomes between the Nuss and Ravitch populations across all age groups, but slightly better outcomes in the Nuss paediatric group as compared to all other groups. Nasr et al, 2010 found no difference in patient satisfaction between both techniques among studies looking at this outcome. A meta-analysis of 2476 cases (1555 Nuss, 921 open surgery) from 23 international studies (Chen et al, 2012) reported more improvement in physiological measures of lung function with the Nuss procedure compared to open surgery, with best results 3 years after surgery. Authors also reported that cardiovascular function after surgery improved by greater than one-half standard deviation. However, no supporting analysis was included in the publication. This meta-analysis was powered to compare physiological pulmonary function change by type of pectus

procedure performed and time after surgery. None of the studies had a healthy (non-pectus) or no-intervention comparator arm or linked the physiological lung function with clinical presentation (dyspnoea, chest pain, exercise intolerance) pre- and post-surgery. Hence, it cannot be used to draw an inference on the clinical effectiveness of pectus procedure on lung function. Authors also reported that cardiovascular function after surgery improved by greater than one-half standard deviation. However, no supporting analysis was included in the publication. Other large case series (Kelly et al, 2013. Žganjer et al, 2011) report positive improvement of chest wall in varying degrees as well as improvement in pulmonary function. Most studies report 80-90% good to excellent anatomic surgical outcomes. Given the limitations in the study design, the overall evidence in this category needs to be viewed with caution.

Outcomes of surgery (infection and revision rates):

There were no studies that directly compared the impact of surgeon volume and outcomes of surgery. In a retrospective review of all primary Nuss procedure repairs of pectus excavatum preformed in a one large US centre over 21 years, complications decreased markedly over 21 years since surgery was first offered in the centre. Bar displacement rate requiring surgical repositioning decreased from 12% in the first decade to 1% in the second decade (Kelly et al, 2010). This provides a limited view of the impact of surgical experience and patient volume on outcomes.

Quality, safety and adverse events associated with surgical correction:

NICE guidance in 2009 (IPG310; 2009) concluded that current evidence on the safety and efficacy of placement of pectus bar for pectus excavatum (also known as MIRPE or the Nuss procedure) is adequate to support its use provided that normal arrangements are in place for clinical governance, consent and audit. It confirmed that placement of pectus bars for pectus excavatum should be carried out only by surgeons with cardiac and thoracic training and experience, who are capable of managing cardiac or liver injury, and where there are facilities for this. The procedure should be carried out only by surgeons with specific training in inserting the device, and they should perform their initial procedures with an experienced mentor. The efficacy and safety of the procedure was based on data from a UK register for 260 patients and multiple case series, small surveys and expert opinion.

The systematic literature review did not find any randomised control trials or high quality meta analysis that could further update the comparative efficacy of different types of surgeries or provide a comparison with a no-intervention group. The best available evidence comes from a systematic review of 39 studies involving 807 adult and 2716 paediatric cases (Johnson et al, 2014) which focused on comparison of the Ravitch, Nuss, and other surgical treatments for pectus excavatum across age groups. The analysis showed that complication rates varied across studies however Nuss and Ravitch procedures were generally safe for paediatric and adult patients with no perioperative mortality reported. Re-operation rates in adults were highest for implant procedures at 18.8% followed by Nuss 5.3% and Ravitch 3.3% but there was no significant difference in re- operation rates in children. Nasr et al, 2010 found that there was no significant difference in overall complication rates between both techniques in the nine studies included in the meta-analysis. Looking at specific complications, postoperative pneumothorax and hemothorax, the rate of reoperation because of bar migration or persistent deformity was significantly higher in the Nuss group. Most case series identified major and minor complications related with the surgery ranging from allergy to nickel (Nuss bars), pneumothorax, hemothorax and pericardial tears in perioperative period to bar displacement and asymmetrical corrections that required re-operations.

Eligibility and thresholds for surgery:

Leading US centres report inclusion criteria for surgery as severe pectus excavatum that fulfils two or more of the following: CT index greater than 3.25, evidence of cardiac or pulmonary compression on CT or echocardiogram, mitral valve prolapse, arrhythmia, or restrictive lung disease (Kelly et al, 2007. Kelly et al, 2010).

Self-perception has been identified as an important element in decision making in pectus surgery. There is significant body image dysmorphia and poor co-relation between objective physiological and perceived impact (mental quality of life and self-esteem) in patients with pectus deformities (Steinman et al, 2011). This highlights the role of psychological evaluation in patient selection and possible need for counselling and management of expectations for patients with exaggerated dysmorphic tendencies.

Evidence indicates that median age for pectus surgery is increasing, with many surgeries in patients above the age of 18 years without any significant difference in outcomes amongst the younger and older patients. (Johnson et al, 2014. Kelly et al, 2010)

3. Research questions

- Impact of surgical correction on cardiorespiratory reserve and functionality for the patient
- Impact of surgeon volume and outcomes of surgery (infection and revision rates)
- Quality, safety and adverse events associated with surgical correction
- Eligibility and thresholds for surgery

4. Methodology

A review of published, peer reviewed literature has been undertaken based on the research questions set out in Section 3 and a search strategy agreed with the lead clinician and public health lead for this policy area. This has involved a PubMed search and search of the Cochrane database for systematic reviews, in addition to review of any existing NICE or SIGN guidance. The evidence review has been independently quality assured.

An audit trail has been maintained of papers excluded from the review on the basis of the inclusion and exclusion criteria agreed within the search strategy. The full list has been made available to the clinicians developing the policy where requested.

5. Results

A detailed breakdown of the evidence is included in the Appendix.

Appendix One

Level	Study design and intervention			Outcomes							Reference	Other		
Level of evidence	Study design	Study size	Intervention	Category	Primary Outcome	Primary Result	Secondary Outcome	Secondary Result	Study Endpoint	Study Endpoint Result	Reference	Complications noted	Benefits noted	Comments
1-	Systematic	Adults Nuss -262 Ravitch (including modified) - 498 Adult surgical implants - 47 Children Nuss - 1500 Ravitch - 1186 Robicsek - 30	Nuss procedure Ravitch procedure Other Procedures	Clinical effectiveness of the intervention	1. Surgery time 2. Use of epidurals 3. Length of Stay (LOS) 4. Complication rates 5. Reoperation rates 6. Outcomes 7. Improvement in cardiac and pulmonary symptoms	ADULTS 1. the Ravitch procedure took longer (191 min) than the Nuss procedure (94 min). The surgical implants took an average of 137.2 min. 2. None of the Ravitch patients received epidurals, while Nuss patients averaged 3 days of epidural use. 3. There was a slight difference in the LOS; 7.3 days for Nuss patients versus 2.9 days for Ravitch, 4.5 days for surgical implant (statistically not different from Ravitch) 4. Complication rates varied greatly among studies, but ranges of all three procedures overlapped. Non-displacement complication rate for the Ravitch procedure (8%) was much lower than the other two procedures (Nuss 21%, Implant 57.4%). The complications with Nuss were mostly minor compared to implants. 5. Reoperation rates were highest for implant procedures Nuss (5.3%) , Ravitch (3.3%) implants procedure (18.8%). 6. Similar outcomes for Ravitch and Nuss. 7. Symptom improvement reported is similar in Nuss and Ravitch. Information was not available for implants. CHILDREN 1. Ravitch procedure took longer than the Nuss or Robicsek procedure. 2. Greater epidural and intravenous analgesia duration in the Nuss procedure (2.5 and 4.1 days respectively) compared to the Ravitch procedure (0.6 and 2.1 days respectively). 3. No significant difference in LOS. 4.The non-displacement complication rates for the Nuss procedure varied much more than for the Ravitch or Robicsek procedures, and there was a tendency for greater non-displacement complication rates for the Nuss and Robicsek procedures than for the Ravitch (averages: 38%, 36%, and 12.5% respectively). 5. No difference in reoperation rates. 6.The Nuss patients (85%, 73-90%) had slightly better excellent outcomes than Ravitch (76%, 68-78%) patients and clearly better excellent outcomes than Robicsek (54%, 38-68%) patients. The good-excellent outcomes, however, were similar across all three procedures (Nuss 95%, Ravitch 96%, Robicsek 89%). 2. All other characteristics were similar. There was no difference in LOS, percentage requiring reoperation, or bar/strut displacement rates– the averages clustered and ranges overlapped for all three procedures outcomes, however, were similar across all three procedures (Nuss 95%, Ravitch 96%, Robicsek 89%). All other characteristics were similar. There was no difference in LOS, percentage requiring reoperation, or bar/strut displacement . There was no significant difference seen in outcome ratings between the Nuss and Ravitch populations across all age groups, but results suggest slightly better outcomes in the Nuss paediatric group as compared to all other groups.	NA	NA	NA	NA	Johnson, William Rainey; Fedor, David; Singhal, Sunil. Systematic review of surgical treatment techniques for adult and pediatric patients with pectus excavatum. J Cardiothorac Surg 2014;217(6):1080-1089.	-	-	This meta analysis allows comparison of the Ravitch, Nuss, and other surgical treatments for pectus excavatum across age groups which is not available in any single good quality study. The key finding of the analysis was that it did not show any linkage between age of operative treatment with outcome. The authors conclude that the Nuss and Ravitch procedures are safe and effective for paediatric and adult patients. They found no clear difference in outcome ratings between the Nuss and Ravitch populations across all age groups, but slightly better outcomes in the Nuss paediatric group as compared to all other groups. Some of the significant limitations to this analysis which limit its generalisability are as follows: 1. The studies included are mostly case series and reports which is reflective of the current level of evidence available for these interventions 2. The analysis does not take into account surgical skill variations amongst individual surgeons, between centres and over time

3	Case series	327	Nuss (bar/minimally invasive) surgery	Clinical effectiveness of the intervention	1. Late complications (>30 days post-op) 2. Reoperation rates 3. Improvement (normalisation) of chest wall 4. Functional Improvement (change in pulmonary function)	1. Of 182 patients with complete follow-up (56%), 18% had late complications, similarly distributed, including substernal bar displacement in 7% and wound infection in 2%. 2. All 13 cases (7%) of bar or strut displacement required reoperation. Data shows another 2 cases of displacement due to trauma. It is not clear if these were included in the 13 cases and whether a reoperation was needed. There were no deaths. 3. 93.8% of patients showed positive improvement of chest wall in varying degrees. Mean initial CT scan index of 4.4 improved to 3.0 post operation (severe >3.2, normal = 2.5). Computed tomography index improved at the deepest point (xiphoid) and also upper and middle sternum. In a small subset of patients (6.2%; n = 10), the pectus index at the deepest point was actually worse after operation, with a mean increase of 0.30 (SD= 0.21). 4. Pulmonary function tests improved (forced vital capacity from 88% to 93%, forced expiratory volume in 1 second from 87% to 90%, and total lung capacity from 94% to 100% of predicted ($p < 0.001$ for each). VO2 max during peak exercise increased by 10.1% (p ¼ 0.015) and O2 pulse by 19% (p ¼ 0.007) in 20 subjects who completed both pre- and postoperative exercise tests.	NA	NA	NA	NA	Kelly, Robert E.; Mellins, Robert B.; Shamberger, Robert C.; Mitchell, Karen K.; Lawson, M. Louise; Oldham, Keith T.; Azizkhan, Richard G.; Hebra, Andre V.; Nuss, Donald; Goretsky, Michael J.; Sharp, Ronald J.; Holcomb, George W.; Shim, Walton K. T.; Megison, Stephen M.; Moss, R. Lawrence; Fecteau, Annie H.; Colombani, Paul M.; Cooper, Dan; Bagley, Traci; Quinn, Amy; Moskowitz, Alan B.; Paulson, James F.. Multicenter study of pectus excavatum, final report: complications, static/exercise pulmonary function, and anatomic outcomes. J. Am. Coll. Surg. 2013;95(3):1043-1049.	Refer to outcome results	Refer to outcome results	The study concludes that there is significant improvement in chest shape, lung function at rest and exercise after surgical correction of pectus excavatum. It also concludes that the surgery can be performed safely in a variety of centres. The key limitation of the study is the absence of a no-intervention control group as well as randomised assignment of treatment options. This is understandably challenging in a practical setting, therefore this case series of fairly large number of patients was shortlisted for clinical evidence review. Patients were scheduled for operative repair by the method of choice for the particular surgeon and family (of 327 patients, 284 underwent Nuss procedure and 43 underwent open procedure). It should be noted that a failure to enrol similar numbers of open and Nuss operation patients also compromised the ability to compare the two operations. There was no stratification of data by surgical team's experience, post-operative care regime etc. Authors have highlighted the difficulty in standardising the exercise data due to reporting protocol and equipment difference. In view of the above and the fact that the study has significant risk of response bias as only 56% of patient (182 out of 327) completed the follow up and that the lung function conclusion was based on a subset of 20 subjects who completed both pre-and postoperative exercise tests, the study results should only be reviewed as low grade clinical evidence for late complications and chest wall normalisation outcomes for Nuss procedure, without weight, age or standardisation for surgical experience and infrastructure.
3	Case series	9	Nuss with open heart surgery	Safety of the intervention	Safety and effectiveness of Nuss when performed with cardiac procedure performed through median sternotomy	Eight patients had bar removal after an average period of 30.3 months. No PE recurrence, bar displacement, or upper sternal depression was reported in 7 patients. Post-operatively, 1 patient exhibited pectus carinatum after a separate spinal fusion surgery for scoliosis. One patient died of unrelated cardiac complications before bar removal.	NA	NA	NA	NA	Sacco Casamassima, Maria Grazia; Wong, Ling Ling; Papandria, Dominic; Abdullah, Fizan; Vricella, Luca A.; Cameron, Duke E.; Colombani, Paul M.. Modified nuss procedure in concurrent repair of pectus excavatum and open heart surgery. Ann. Thorac. Surg. 2013;2013(0):850840.	NA	NA	Very small study (9 patients) of patients with severe pectus excavatum (PI>3) with congenital and acquired cardiac disorders that also require surgical repair. A case series with obvious selection bias and lack of inclusion controls despite a potential cohort (29 patients with severe PE who underwent cardiac surgery either in a single or staged approach) being available in the initial selection patients.

1+	Other	2476 patients with PE, including 921 who underwent open surgical repair (Ravitch + Daniel) and 1555 who underwent the minimally invasive Nuss technique	Nuss procedure	Clinical effectiveness of the intervention	Change in pulmonary function (forced expiratory volume over 1 second (FEV1), forced vital capacity (FVC), vital capacity (VC), and total lung capacity (TLC) following surgical repair	FEV1 increased significantly 3 years after surgical correction of PE using a minimally invasive technique, with the Nuss procedure associated with better results than the Ravitch procedure. (FEV1 changes after surgical correction favoured the Ravitch procedure at 1 year (WMD = 2.19, 95%CI −4.18 ~ 8.56). But favoured the Nuss procedure at 3 years (WMD = 3.00, 95%CI −0.47 ~ 6.46). Although FVC decreased within 1 year after surgical correction of PE using both minimal and open techniques, greater FVC improvement occurred 3 years after the Nuss (WMD = 4.31, 95%CI −1.80 ~ 10.42) than after the Ravitch (WMD = 0.28, 95%CI −0.15 ~ 0.41) procedure and bar removal. VC changes favoured the Ravitch procedure postoperatively (WMD = 4.34, 95%CI −4.31 ~ 12.98), but three years after VC increased significantly, with better results for Nuss (WMD = 3.52, 95%CI −2.44 ~ 9.49) than for the Ravitch procedure (WMD = 0.05, 95%CI −0.07 ~ 0.16) procedure. TLC changes after one year favoured Nuss correction preoperatively (WMD = −3.96, 95%CI −11.75 ~ 3.82).Three years after surgery, TLC improved after the Nuss procedure, showed better postoperative results (WMD = 3.52, 95%CI −3.87 ~ 4.20) than the Ravitch procedure (WMD = 0.18, 95%CI 0.06 ~ 0.31).	1. Improvement in cardiovascular function 2. Ideal age to operate	1. Detailed analysis of this was not reported but the authors discuss that cardiovascular function after surgery improved by greater than one-half standard deviation, supporting the hypothesis that relief of cardiac compression caused by the depressed sternum improves the hemodynamic responses of patients with PE 2. Authors have made observations about age-related outcomes but we were unable to find studies with similar finding in the meta-analysis	Long-term and sustained improvement in PE symptoms and presentation	NA	Chen, Zhenguang; Amos, Ela Bella; Luo, Honghe; Su, Chunhua; Zhong, Beilong; Zou, Jianyong; Lei, Yiyan. Comparative pulmonary functional recovery after Nuss and Ravitch procedures for pectus excavatum repair: a meta-analysis. J Cardiothorac Surg 2012;23(4):486-491.	NA	NA	This meta-analysis was powered to compare physiological pulmonary function change by type of pectus procedure performed and time after surgery. Hence, it cannot be used to draw an inference on the clincially significant effectiveness of pectus procedure on lung function. None of the studies had a healthy (non-pectus) or no-intervention comparator arm or linked the physiological lung function with clinical presentation (dyspnoea, chest pain, exercise intolerance) pre- and post-surgery. The authors note that while the lung function improvement may be relevant to patients with severe deformities, most studies included in the meta-analysis showed only modest preoperative reduction in vital capacity and total lung capacity prior to surgery which may not be clinically relevant.
3	Case series	128	Nuss	Clinical effectiveness of the intervention	1. Perioperative features: Length of stay, epidural duration 2. Complications 3. Outcomes	Length of hospital stay: 7 to 24 days (average 10 days). Epidural: 3 to 6 days (average 4 days) Complications: Operative mortality: zero. All patients had pneumothorax in the course of operation treatment because surgeons used thoracoscopy with CO_2 insufflations. Post-operative complications included 36 pneumothorax, two clinically insignificant pericardial tears without other complications, one patient had a fracture of the sternum, six had pneumonia, two developed pericarditis, one patient had hematothorax, two had bar infections and two had cellulitis. Outcome: Early results: Excellent results (75%), good result in 25 (19%) and poor (6%) of patients. Longer term results: The same results were maintained over the follow-up period which was between six months and five years (mean 3.6 years). Authors report significant improvement in clinical symptoms especially shortness of breath, chest pain on exercise and shift and compression of heart in 69 patients for whom they conducted a pre and post evaluation.	Outcome of bar removal	After bar removal in 74 patients, 54 (72.9%) had excellent results and maintained a normal chest. Good results were maintained in 16 patients (21.6%) with mild residual pectus and poor results in four patients (5.5%) with severe recurrence.	NA	NA	Žganjer, Mirko; Žganjer, Vlasta. Surgical correction of the funnel chest deformity in children. Int Orthop 2011;20(4):583-597.	Refer to outcome results	Refer to outcome results	This was a case series of children - there were cardiac and pulmonary problems in 69 (54%) patients, and 59 (46%) patients had cosmetic and cosmetic-related psychological problems, progression of the deformity with other symptoms and Haller index greater than 3.25. The authors conclude that the Nuss procedure is an effective method with excellent cosmetic results, low percentage of complications and excellent improvement in cardiopulmonary status. This is a case series study of cases performed in a single centre possibly by the authors. With the inherent bias in the study due to its design methodology and the absence of standardised measure/scale to weigh clinical benefits (physical, psychological and quality of life) against the significant morbidity caused by the procedure, the author's conclusion on effectiveness of the surgery appear unsupported by good quality evidence.

2-	Other	NA	Nuss Procedure	Clinical effectiveness of the intervention	Perioperative outcomes	There was no significant difference in overall complication rates between both techniques (OR, 1.75 (0.62-4.95); P = .30). Looking at specific complications, the rate of reoperation because of bar migration or persistent deformity was significantly higher in the Nuss group (OR, 5.68 (2.51-12.85); P = .0001). Also, post-operative pneumothorax and hemothorax were higher in the Nuss group (OR, 6.06 [1.57-23.48]; P = .009 and OR, 5.60 [1.00-31.33]; P = .05), respectively. Duration of surgery was longer with the Ravitch (WMD, 69.94 minutes (0.83-139.04); P = .05). There was no difference in length of hospital stay (WMD, 0.4 days (-2.05 to 2.86); P = .75) or time to ambulation after surgery (WMD, 0.33 days [-0.89 to 0.23]; P = .24). Among studies looking at patient satisfaction, there was no difference between both techniques. Our results showed no difference between both techniques with regard to overall complication rates (OR, 1.75 [0.62-4.95]; P = .30) (Fig. 1). Specific complications such as the rate of reoperation after the Nuss procedure was higher compared to the Ravitch (OR, 5.68 [2.51-12.85]; P = .0001) (Fig. 2). The indication for reoperation was either for persistent deformity or bar migration. Stabilizers applied to the Nuss bar to prevent bar migration were not used in some patients in the included studies; however, because of data availability, we were not able to perform a subgroup analysis in these patients to determine the importance of the stabilizers in preventing reoperation. Also, postoperative pneumothorax and hemothorax were higher in the Nuss group. There was no difference about blood transfusion requirement. The duration of surgery was longer in the Ravitch group by almost 70 minutes (WMD, 69.94 minutes [0.83-139.04]; P = .05) (Fig. 3). There was no difference with regard to the length of hospitalization (WMD, 0.4 days [−2.05 to 2.86]; P = .75) (Fig. 4) or time to ambulation (WMD, 0.33 days [0.89-0.23]; P = .24; Fig. 5), between both groups.	NA	NA	NA	NA	Nasr, Ahmed; Fecteau, Annie; Wales, Paul W.. Comparison of the Nuss and the Ravitch procedure for pectus excavatum repair: a meta-analysis. J. Pediatr. Surg. 2010;88(6):1773-1779.	Refer to outcome results	Refer to outcome results	The study is a systematic review and limited meta analysis of the data from 9 retrospective/prospective (case series) studies. The search methodology used by authors conforms more with finding comparable studies for inclusion in a meta analysis instead of a complete systematic review on the topic. The authors found no randomised control trials fit for inclusion on systematic literature search. We therefore agree with the authors that while the results of this meta-analysis fail to provide overwhelming support to either approach, and both approaches are acceptable. Meta analysis for observational studies, such as this one, has the risk of significant contamination from bias, confounding and statistical heterogeneity between studies.
3	Case-control	40	Nuss	Clinical effectiveness of the intervention	Perioperative outcomes	The time of surgery was greater with SCP than with Nuss and epidural was longer for Nuss. There was no difference in relation to duration of hospital stay and follow-up. No significant differences were found when comparing the number of patients who had complications between the two groups. However, when comparing the number of complications in each group, more complications were found in the Nuss group. More positive results were observed in the SCP group than the Nuss group but the difference was not statistical significant. Most patients in both groups had favourable results and were very satisfied with the aesthetic results achieved.	-	-	-	-	Coelho, Marlos de Souza; Silva, Ruy Fernando Kuenzer Caetano; Bergonse Neto, Nelson; Stori, Wilson de Souza; dos Santos, Anna Flávia Ribeiro; Mendes, Rafael Garbelotto; Fernandes, Lucas de Matos. Pectus excavatum surgery: sternochondroplasty versus Nuss procedure. Ann. Thorac. Surg. 2009;44(5):888-892.	Refer to outcome results	Refer to outcome results	This is not a case control study but an observation of outcomes of a retrospectively selected cohort of patients. The criteria for selection of patients in the study and for the type of surgery is unclear. In view of the study design and analysis undertaken, the authors' conclusion that Sternochondroplasty surgery yielded better results than the Nuss procedure for asymmetric pectus excavatum is evidenced only for the 40 patients included in the study.

3	Case series	327	Nuss (bar/minimally invasive) surgery	Clinical effectiveness of the intervention	A. Is anatomically severe pectus excavatum associated with abnormal pulmonary function? B. Early results (up to 3 months post operative) 1. Length of stay 2. Perioperative complications and mortality 3. Pain, different between two procedures	Because of disproportionate enrolment and similar early complication rates, statistical comparison between operation types was limited. A. Median preoperative CT index was 4.4. Pulmonary function testing before operation showed mean forced vital capacity of 90% of predicted values; forced expiratory volume in 1 second (FEV(1)), 89% of predicted; and forced expiratory flow during the middle half of the forced vital capacity (FEF(25% to 75%)), 85% of predicted. B. Early post correction results showed that operations were performed without mortality and with minimal morbidity at 30 days postoperatively. Median hospital stay was 4 days. C. Post-operative pain was a median of 3 on a scale of 10 at time of discharge; the worst pain experienced was the same as was expected by the patients (median 8), and by 30 days after correction or operation, the median pain score was 1.	NA	NA	NA	NA	Kelly, Robert E.; Shamberger, Robert C.; Mellins, Robert B.; Mitchell, Karen K.; Lawson, M. Louise; Oldham, Keith; Azizkhan, Richard G.; Hebra, Andre V.; Nuss, Donald; Goretsky, Michael J.; Sharp, Ronald J.; Holcomb, George W.; Shim, Walton K. T.; Megison, Stephen M.; Moss, R. Lawrence; Fecteau, Annie H.; Colombani, Paul M.; Bagley, Traci C.; Moskowitz, Alan B.. 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;16(6):639-642.	Refer to outcome results	Refer to outcome results	The study is part 1 of 2, reporting the early findings by the same authors (Kelly et al 2013). The study concludes that early post correction results showed that operations were performed without mortality and with minimal morbidity at 30 days postoperatively. The safety and the acceptability of the perioperative complications in a group of younger patients has to be weighted with the real need of surgical intervention (principle of clinical equipoise). In this respect, the key limitation of the study remains the absence of a no-intervention control group, non- randomised assignment of treatment options, limitations in comparing data by type of procedures (majority of patients had Nuss intervention) and by surgical skill. For additional comments, refer to Kelly et al 2013.

2-	Case-control	90 cases; 82 controls	Quality of life and body image prior to surgical correction in pectus deformity patients	Other	1. Objective severity of the deformity: funnel-chest index by Hümmer and the Haller index). 2.Quality of life 2.a. Disease-specific quality of life(Nuss Questionnaire modified for Adults (NQ-mA) 2.b. Health-related quality of life was determined by the Short-Form-36 Health Survey (SF-36) 3. Body Image 3.a. Body Image Questionnaire (FKB-20) 3.b. Dysmorphic Concern Questionnaire (DCQ) 3.c. Self-evaluation of the subjective impairment of the appearance 4. Psychological Health 4.a. Diagnostic Interview for Mental Disorders – Short Version (Mini-DIPS) 4.b. General Depression Scale (ADS) 4.c. Self-rating of self-esteem	Compared with control group results, physical quality of life was reduced in patients with pectus excavatum, while mental quality of life was decreased in patients with pectus carinatum ($p<0.05$). Body image was highly disturbed in all the patients and differed significantly from the control group ($p<0.01$). Patients with pectus carinatum appeared to be less satisfied with their appearance than those with pectus excavatum ($p=0.07$). Body image distress was multi-variately associated with both reduced mental quality of life and low self-esteem ($p<0.001$). Body image did not influence physical quality of life. Patients displayed no elevated rates of mental disorders according to Diagnostic and Statistical Manual of Mental Disorders, fourth edition (DSM-IV) criteria.	NA	NA	-	-	Steinmann, Cornelia; Krille, Stefanie; Mueller, Astrid; Weber, Peter; Reingruber, Bertram; Martin, Alexandra. 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;25(5):421-424.	-	-	Self-perception is an important element in decision making in pectus surgery. This study provides a framework of body image evaluation that could be included in the assessment of patients with chest deformities. Additional post-surgical assessment would have been useful in evaluating the impact of surgical correction on quality of life and body image especially in patients with exaggerated dysmorphic concerns. There is significant evidence in literature on limited satisfaction with the surgical outcome in patients with extreme dysmorphia. The key limitation of the study is the possibility of selection bias given that only patients with pectus deformity who presented themselves for surgical intervention were included in the study. Additionally, it is a retrospective case control study with significant hetrogenity within subgroups including difference in study population numbers , has not been adequately addressed.
3	Case series	69	Conservative treatment with custom-fitted brace, worn 12-15 hours a day for up to 1 year.	Clinical effectiveness of the intervention	1. Correction angle of PC measured by standardised lateral views 2. Patient rated outcomes	1. Mean correction angle of 10 degrees in the children's group and 5 degrees in the adolescent group 2. 82% of adolescent patients judged the result as "excellent" or "good"	Impact of compliance	Patients who reported the result "unchanged" had a mean daily brace wearing time of 8.73 hours, those who judged the result as "good" 14.53 hours, and those who judged the result as "excellent" 18.36 hours	-	-	Loff, Steffan; Sauter, Hartwig; Wirth, Thomas; Otte Ralf. Highly Efficient Conservative Treatment of Pectus Carinatum in Compliant Patients. Eur J Pediatr Surg 2015;96(1):272-278.	-	-	This is a single institution case series involving a selective group of patients. While the results look promising, absence of randomised selection of patients, comparator group and long term follow-up on patients (reversion of bone deformities after removal of external support is known), limit the wider application of the findings.

2-	Case-control	49 patients 26 controls	Lung function and cardiac index at rest and bicycle exercise, before, 1 year and 3 years after Nuss procedure for pectus excavatum (PE)	Clinical effectiveness of the intervention	1. Lung function 2. Cardiac Index	1. Preoperatively, patients had lower forced expiratory volume in the first second of expiration (FEV1; 86% ± 13%) as compared with controls (94% ± 10%), p = 0.009. Postoperatively, no difference was found in FEV1 between the 2 groups. 2. Preoperatively, patients had lower maximum cardiac index, mean ± SD, 6.6 ± 1.2 l·min(-1)·m(-2) compared with controls 8.1 ± 1.0 l·min(-1)·m(-2) during exercise (p = 0.0001). One year and 3 years postoperatively, patients' maximum cardiac index had increased significantly and after 3 years there was no difference between patients and controls (8.1 ± 1.2 l·min(-1)·m(-2) and 8.3 ± 1.6 l·min(-1)·m(-2), respectively [p = 0.572]).	NA	NA	-	NA	Maagaard, Marie; Tang, Mariann; Ringgaard, Steffen; Nielsen, Hans Henrik M.; Frøkiær, Jørgen; Haubuf, Maj; Pilegaard, Hans K.; Hjortdal, Vibeke E.. Normalized cardiopulmonary exercise function in patients with pectus excavatum three years after operation. Ann. Thorac. Surg. 2013;252(6):1072-1081.	-	-	While this study shows the difference in FEV1 and cardiac index for pectus patients compared to normal adults, actual clinical significance of the comparative reduction in FEV1 and cardiac index preoperatively and the improvement post-operatively was not established. In addition, the potential for selection bias and hence the representativeness of case and control groups has not been adequately addressed.
3	Case series	2378 (1215 of these patients underwent surgery)	NUSS procedure	Clinical effectiveness of the intervention	-	In primary operation: 1 bar was placed in 69%, 2 bars in 30% and 3 bars in 0.4% Perioperative Complications: Allergy to nickel: 28% (35 patients) Wound infection: 4% (17 patients) Hemothorax: 0.6% (8 patients) Volume and learning curve Complications decreased markedly over 21 years since surgery was first offered in the centre. Bar displacement rate requiring surgical repositioning decreased from 12% in the first decade of the surgeries to 1% in the second decade. The median age of patients has gradually shifted over the years from 6 years to 14 years with up to 10% of patients above 18 years of age.	Post-operative outcomes	A good or excellent anatomic surgical outcome was achieved in 95.8% of patients at the time of bar removal. A fair result occurred in 1.4%, poor in 0.8%, and recurrence of sufficient severity to require reoperation occurred in 11 primary surgical patients (1.4%).	-	-	Kelly, Robert E.; Goretsky, Michael J.; Obermeyer, Robert; Kuhn, Marcia Ann; Redlinger, Richard; Haney, Tina S.; Moskowitz, Alan; Nuss, Donald. Twenty-one years of experience with minimally invasive repair of pectus excavatum by the Nuss procedure in 1215 patients. Ann. Surg. 2010;148(2):657-661.	-	-	This is a single institution case series. All patients have been included in the study. Comparison of outcomes amongst surgeons is not included. The study provides a view of the impact of surgical experience and patient volume on outcomes.

Appendix Two

Literature search terms

Assumptions / limits applied to search:	
Original search terms:	The search will look at the NUSS and Ravitch operations rather than minor procedures. The search will also consider the paediatric and adult populations separately.
Updated search terms - Population	Pectus Deformities Pectus Carinatum Currarino-Silverman Syndrome Pectus Carinatum, Arcuate Pectus Carinatum, Chondrogladiolar Pectus Carinatum, Chondromanubrial Pouter Pigeon Breast Pectus Excavatum Funnel Chest Pectus Abnormalities Sternum Abnormalities pectus anomaliesas, scoliosis, marfans syndrome
Updated search terms - Intervention	Mirpe Procedure Nuss Procedure Ravitch Procedure
Updated search terms - Comparator	Subcutaneous Implant Breast Augmentation Suction Devices Minor Surgery Minor Surgeries
Updated search terms - Outcome	None
Inclusion criteria	General inclusion criteria
	In order of decreasing priority, the following are included: 1. All relevant systematic reviews and meta-analysis in the last 5 years and those in 5-10 years period which are still relevant (e.g. no further updated systematic review available) 2. All relevant RCTs and those in the 5-10 years period which are still relevant (e.g. not superseded by a next phase of the trial / the RCT is one of the few or only high quality clinical trials available) >>>> If studies included reach 30, inclusion stops here 3. All relevant case control and cohort studies, that qualify after exclusion criteria >>>> If studies included reach 30, inclusion stops here 4. All relevant non analytical studies (case series/ reports etc.) that qualify after exclusion criteria >>>> If studies included reach 30, inclusion stops here 5. Expert opinion

	Specific inclusion criteria
	English language Published after 2009 (following the NICE guidelines published in the same year) Title/Abstract The PICO specifies a distinction between adult and paediatric evidence, although age filters are not applied in the first instance due to inconsistent results. 4 additional articles as per the suggestion of the Policy Working Group: a. Kelly, Robert E.; Shamberger, Robert C.; Mellins, Robert B.; Mitchell, Karen K.; Lawson, M. Louise; Oldham, Keith; Azizkhan, Richard G.; Hebra, Andre V.; Nuss, Donald; Goretsky, Michael J.; Sharp, Ronald J.; Holcomb, George W.; Shim, Walton K. T.; Megison, Stephen M.; Moss, R. Lawrence; Fecteau, Annie H.; Colombani, Paul M.; Bagley, Traci C.; Moskowitz, Alan B.. 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;16(6):639-642. b. Coelho, Marlos de Souza; Silva, Ruy Fernando Kuenzer Caetano; Bergonse Neto, Nelson; Stori, Wilson de Souza; dos Santos, Anna Flávia Ribeiro; Mendes, Rafael Garbelotto; Fernandes, Lucas de Matos. Pectus excavatum surgery: sternochondroplasty versus Nuss procedure. Ann. Thorac. Surg. 2009;44(5):888-892. c. Loff, Steffan; Sauter, Hartwig; Wirth, Thomas; Otte, Ralf. Highly Efficient Conservative Treatment of Pectus Carinatum in Compliant Patients. Eur J Pediatr Surg 2015;96(1):272-278. d. Steinmann, Cornelia; Krille, Stefanie; Mueller, Astrid; Weber, Peter; Reingruber, Bertram; Martin, Alexandra. 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;25(5):421-424.
Exclusion criteria	**General exclusion criteria**
	Studies with the following characteristics will be excluded: 1. Do not answer a PICO research question 2. Comparator differs from the PICO 3. < 50 subjects (except where there are fewer than 10 studies overall) 4. No relevant outcomes 5. Incorrect study type 6. Inclusion of outcomes for only one surgeon/doctor or only one clinical site
	Specific exclusion criteria
	• Mild pectus deformities • Age. The majority of patients undergoing treatment are between 14-18 years of age. There is some variation in practice between paediatric and adult thoracic surgeons regarding age but most thoracic surgeons tend not to offer surgery (for PE) after 30 years of age. Technically, though, there is no age restriction.