NHS England

Evidence review: Surgical correction for pectus deformity

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Contents

1 Introduction

- Pectus abnormalities cover a range of deformities affecting the anterior chest wall, specifically the sternum and adjacent rib cartilages. Pectus excavatum (PE) and pectus carinatum (PC) are the most common of these.
- PE or funnel chest appears as a depression of the sternum; it may be asymmetrical, with the right side deeper than the left (de Oliveira Carvalho, 2014). It can be present at birth or may develop during childhood and adolescence. PC, also known as pigeon chest, is caused by the sternum pushing out so the middle of the chest is more pronounced (Goretsky et al 2004). It usually manifests at the time of a growth spurt in the early teenage years (Fokin et al 2009).
- Both deformities may be inherited, often being present in several members of the same family and associated with other congenital diseases including scoliosis of the spine, Marfan's syndrome and Ehlers-Danlos syndrome (Goretsky et al 2004).
- Birth incidence is between 1 in 400 and 1 in 1000, of which 87% will be PE, 5% PC and the remainder a combination of the two or other very rare chest deformities (Lomholt et al 2016, Krasopoulos et al 2010, Goresky et al 2004). Both PE and PC are more likely to present in males, with a male to female ratio of between 3:1 and 9:1 (Krasopoulos et al 2010, Goresky et al 2004, Kelly et al 2004).
- Pectus deformities vary from mild and asymptomatic to severe, impacting on pulmonary and cardiac function. Diagnosis is by physical examination (Lain et al 2017). Psychologically, the deformity usually has little impact, but in moderate and severe cases it can cause significant psychological distress leading to concerns about appearance, withdrawal and social isolation (Steinman et al 2011).
- PE is surgically repaired using two techniques. The Nuss procedure (also known as minimally invasive repair of pectus excavatum – MIRPE) is generally only applicable in PE whilst the Ravitch procedure can be used for both PE and PC. The Nuss procedure involves placing one or two steel bars under the breastbone with the aim of raising it and correcting the abnormal shape. Each bar, bent into a curve to fit the patient's chest, is inserted through small openings in the chest. The bar (or bars) is/are usually removed within a few years of placement. In the modified Ravitch procedure, the rib cartilages are cut away on each side and the sternum is flattened so that it will lie flat. One or more permanent bars or struts are inserted to ensure the sternum keeps its shape.
- Treatment is determined by assessment of the type of pectus deformity, degree of deformity, simple versus mixed deformity, and determination of whether the deformity is isolated or part of a syndrome**.** The severity of the deformity can be assessed radiologically using the Haller index, defined as the lateral external distance divided by the distance between the external deepest point of the chest and external spinous process.
- The surgical treatment of pectus deformities falls within the commissioning responsibilities of NHS England. Between 300 and 500 operations to correct pectus deformity are carried out in England each year (NHS England 2016), covering only a minority of people with the deformity. Non-surgical options include posture and exercise programmes and bracing.
- NICE concluded that that evidence on the safety and efficacy of MIRPE or the Nuss procedure is adequate to support its use provided that normal arrangements are in place for clinical governance, consent and audit (NICE 2009).

2 Summary of results

- Six papers matching the PICO were included in this review. Four reported results in a cohort of participants before and after surgery (Luo et al 2017, Lomholt et al 2016, Kuru et al 2015 and Kelly et al 2008). Two reported results in participants after surgery for pectus deformity: Bahadir et al 2017 compared a group of patients with pectus deformity who had surgery with a group who had not had surgery; Jacobsen et al 2011 compared patients who had surgery for pectus deformity with controls who had no pectus deformity. The total number of participants with pectus deformity included in the studies was 907 (range 63 to 266) and ages of participants across the studies ranged from 8 to 29 years. None of the studies was randomised.

- Five studies reported results about social functioning and emotional or behavioural problems (Luo et al 2017, Lomholt et al 2016, Kuru et al 2015, Kelly et al 2008 and Jacobsen et al 2010).
    - Luo et al 2017 (n=266) reported that there was no statistically significant improvement in interpersonal sensitivity after surgery.
    - Lomholt et al 2016 (n=107) reported that patients' mean 'role/social limitations: behavioural' scores improved from 94.8/100[1] before surgery to 99.2/100 six months after surgery (p=0.004). They also reported that patients' mean scores for 'role/social limitations: emotional' improved from 90.6/100 to 98.7/100 (p<0.0001) over the same period. In this study, parents' mean combined scores for 'role/social limitations: emotional and behavioural' improved from 89.6/100 before surgery to 98.5/100 six months after (p=0.001).
    - Kuru et al 2015 (n=88) compared psychosocial functioning before and six months after surgery. Patients' median scores improved from 22.5/48 to 33/48 (p=0.000), and parents' scores from 20/44 to 24/44 (p=0.000).
    - Kelly et al 2008 (n=264) reported that parents' mean scores[2] for the emotional difficulties of their child improved from 1.81/4 before surgery to 1.24/4 a year after (p<0.0001). Their mean scores for their child's social self-consciousness also improved, from 2.86/4 to 1.33/4 (p<0.0001).
    - Lastly, Jacobsen et al 2010 (n=119) reported higher mean scores[3] for 'role/social limitations: emotional' in patients who had had surgery for pectus deformity (96.7/100) than in age-matched healthy controls (90.6/100, p<0.001). A similar result was reported for role/social limitations: behavioural (98.2/100 vs 95.4/100 respectively, p<0.001).

- Mental health was reported in four studies (Luo et al 2017, Lomholt et al 2016, Jacobsen et al 2010 and Bahadir et al 2017).
    - Luo et al 2017 reported a fall in the proportion of participants above a threshold for the diagnosis of mental health problems (preoperative 161/266 (60.5%), postoperative 79/266 (29.7%), p < 0.001), along with a similar fall in the proportion of participants above a threshold for the diagnosis of depression (preoperative 153/266 (57.5%), postoperative 76/266 (28.6%), p < 0.001). However, the levels of patients' anxiety scores were not altered after surgery in this study.
    - Lomholt et al 2016 reported an improvement in patients' mean mental health

[1] This indicates that the measurement scale has a maximum score of 100, where 0 equals worst health and 100 equals 'best health'.
[2] Out of a maximum score of 4, where lower scores (for both parent and child questionnaires) mean better health status and 1 = 'very happy' .
[3] Out of a maximum score of 100, where 0 equals worst health and 100 equals best health .

scores: before surgery, the mean score was 82.6/100, whereas six months after surgery it was 86.2/100 (p=0.04). Parents also reported a rise in their child's mental health score, from 84.9/100 to 87.9/100 over the same period (p=0.04).
    - Jacobsen et al 2010 reported a better mental health score in patients who had had surgery for pectus deformity (83.7/100) than in age-matched healthy controls (78.4/100) (p<0.001.
    - Bahadir et al 2017 (n=63) reported no significant difference in anxiety scores and depression scores between patients with pectus deformity after surgery and those who had no surgery.
- Kelly et al 2008 reported that patients' mean body image scores[4] improved from 2.30/4 before surgery to 1.40/4 a year after surgery, (p<0.0001), a lower score indicating better body image.
- Luo et al 2017 reported an improvement of mean somatisation score[4] from 1.57/5 before surgery to 1.23/5 a year after, an improvement of 0.34/5 (p = 0.001).
- Lomholt et al 2016 reported an improvement in patients' mean self-esteem scores from 83.0/100 before surgery to 89.3/100 six months after (p<0.0001). Parents also rated their child's self-esteem as improved, from 77.0/100 to 83.7/100 over the same period.
- Lomholt et al 2016 reported that patients' mean behaviour scores improved from 87.4/100 before surgery to 89.9/100 six months after surgery (p=0.05). Parents' mean scores for their child's behaviour did not improve significantly.
- Lomholt et al 2016 reported that patients' mean family activity scores improved from 88.0/100 before surgery to 95.2/100 six months after surgery (p=0.001). Parents' mean scores for family cohesion also improved from 89.0/100 to 95.2/100 over the same period (p<0.0001). Patients reported no change in family cohesion, but parents' mean scores improved from 79.1/100 to 84.8/100 over the six months (p=0.03). Jacobsen et al 2010 reported better family activities scores in patients who had had surgery for pectus deformity (90.6/100) than in age-matched healthy controls (82.6/100) (p<0.001).
- No evidence was found about the relationship between degree of the pectus deformity and change in psychological outcomes, about sub-groups in which surgery produces a greater improvement in mental wellbeing or about whether incorporation of psychosocial therapies into surgical management affects outcome.
- There are a number of serious weaknesses in the evidence that was found. The studies were not well-controlled. The studies provide little information on the significance to patients of the differences that they found. In many cases, patients' and parents' scores did not indicate the existence of major psychosocial or other difficulties before surgery, limiting the improvement that surgery could provide. The outcome measures varied between studies and were often opaque. There were many important data the authors did not report. The involvement of parents in the studies may have introduced bias into the studies' results.
- It is unclear the extent to which the changes reported are attributable to surgery and whether they are large enough to make a difference to patients. There is a need for better reported studies with more appropriate controls before one can be confident about the effect of surgery for pectus deformities.

[4] Higher scores indicate worse symptoms

3 Methodology

- The methodology to undertake this review is specified by NHS England in their 'Guidance on conducting evidence reviews for Specialised Commissioning Products' (2016).
- A description of the relevant Population, Intervention, Comparison and Outcomes (PICO) to be included in this review was prepared by NHS England's Policy Working Group for the topic (see section 9 for PICO).
- The PICO was used to search for relevant publications in the following sources: PubMed, Embase, Cochrane Library and PsychINF (see section 10 for search strategy).
- The search dates for publications were 10th November 2007 to 9th November 2017.
- The titles and abstracts of the results from the literature searches were assessed using the criteria from the PICO. Full text versions of papers which appeared potentially useful were obtained and reviewed to determine whether they were appropriate for inclusion. The higher quality papers which matched the PICO criteria were then selected for inclusion in this review; studies with fewer than fifty participants were excluded because larger studies were found. Studies which reported patient satisfaction as the only psychological outcome were excluded from the review.
- Evidence from all papers included was extracted and recorded in evidence summary tables, critically appraised and their quality assessed using the National Service Framework for Long Term Conditions (NSF-LTC) evidence assessment framework (see section 7).
- The body of evidence for individual outcomes identified in the papers was graded and recorded in grade of evidence tables (see section 8).

4 Results

Six papers matching the PICO were included in this review. Four reported results in a cohort of participants before and after surgery (Luo et al 2017, Lomholt et al 2016, Kuru et al 2015 and Kelly et al 2008). Two reported results in participants after surgery for pectus deformity: Bahadir et al 2017 compared a group of patients with pectus deformity who had surgery with a group who had not had surgery; Jacobsen et al 2011 compared patients who had surgery for pectus deformity with controls who had no pectus deformity. The total number of participants with pectus deformity included in the studies was 907 (range 63 to 266). None of these studies was randomised.

The age ranges of participants in these studies were as follows: Luo et al 2017 mean 19.02 years, standard deviation 4.42 years; Lomholt et al 2016, 11 to 20 years; Kuru et al 2015, 14 to 29 years; Kelly et al 2008, 8 to 21 years; Bahadir et al 2017, 9 to 17 years; Jacobsen et al 2011, 8 to 20 years.

Full details of the study designs and outcomes are summarised in the evidence tables in section 7, together with details of the outcome measurement scales used.

Does surgical intervention with and without accompanying psychological intervention improve the psychological well-being and quality of life for individuals with pectus deformities?

All six studies reported the results of questionnaires of participants who had undergone surgery,

and four also reported questionnaire results from parents (Lomholt et al 2016, Kuru et al 2015, Kelly et al 2008, and Bahadir et al 2017). The questionnaires assessed aspects of psychological and health-related quality of life (HRQOL) including social functioning, mental health, body image and somatisation, emotional problems, self-esteem, behaviour, family activities and cohesion.

Section 5 includes discussion of the clinical significance of the reported differences, and details of the questionnaires are in section 7.

Social functioning and emotional or behavioural problems

Five studies reported results about social functioning and emotional or behavioural problems (Luo et al 2017, Lomholt et al 2016, Kuru et al 2015, Kelly et al 2008 and Jacobsen et al 2010).

Luo et al 2017 reported that there was no statistically significant improvement in interpersonal sensitivity after surgery.

Lomholt et al 2016 reported that patients' mean 'role/social limitations: behavioural' scores improved from 94.8/100 before surgery to 99.2/100 six months post-surgery (p=0.004). They also reported that patients' mean scores for 'role/social limitations: emotional' improved from 90.6/100 to 98.7/100 (p<0.0001) over the same period. In this study, parents' combined mean scores for 'role/social limitations: emotional and behavioural' improved from 89.6/100 before surgery to 98.5/100 six months after (p=0.001).

Kuru et al 2015 compared psychosocial functioning before and six months after surgery. Patients' median scores improved from 22.5/48 to 33/48 (p=0.000), and parents' scores from 20/44 to 24/44 (p=0.000).

Kelly et al 2008 reported that parents' mean scores for the emotional difficulties of their child improved from 1.81/4 before surgery to 1.24/4 a year after (p<0.0001). Their mean scores for their child's social self-consciousness also improved, from 2.86/4 to 1.33/4 (p<0.0001).

Lastly, Jacobsen et al 2010 reported higher mean scores for 'role/social limitations: emotional' in patients who had had surgery for pectus deformity (96.7/100) than in age-matched healthy controls (90.6/100) (p<0.001). A similar result was reported for 'role/social limitations: behavioural' (98.2/100 vs 95.4/100 respectively, p<0.001).

Mental Health

Mental health was reported in four studies (Luo et al 2017, Lomholt et al 2016, Jacobsen et al 2010 and Bahadir et al 2017).

Luo et al 2017 reported a fall in the proportion of participants above a threshold for the diagnosis of mental health problems (preoperative 161/266 (60.5%), postoperative 79/266 (29.7%), p < 0.001), along with a similar fall in the proportion of participants above a threshold for the diagnosis of depression (preoperative 153/266 (57.5%), postoperative 76/266 (28.6%), p < 0.001). However, the levels of patients' anxiety scores were not altered after surgery in this study.

Lomholt et al 2016 reported an improvement in patients' mean mental health scores. Before surgery, the mean score was 82.6/100, whereas six months after surgery it was 86.2/100 (p=0.04). Parents also reported a rise in their child's mental health score, from 84.9/100 to 87.9/100 over the same period (p=0.04).

Jacobsen et al 2010 reported a better mental health score in patients who had had surgery for pectus deformity (83.7/100) than in age-matched healthy controls (78.4/100) (p<0.001).

Bahadir et al 2017 reported no significant difference in anxiety scores and depression scores between patients with pectus deformity after surgery and others who had not had surgery for their deformity. These authors also reported no significant differences in patient-rated total score, emotional problems, conduct problems, hyperactivity/inactivity, peer relationship problems and prosocial behaviour; there were also no significant differences in parent-rated emotional symptoms, conduct problems, hyperactivity/inactivity, peer relationship problems and pro-social behaviour.

Body image and somatisation
Kelly et al 2008 and Luo et al 2017 reported body image and somatisation scores.

Kelly et al 2008 reported that patients' mean body image scores improved from 2.30/4 before surgery to 1.40/4 a year after surgery, ($p<0.0001$), a lower score indicating better body image. Luo et al 2017 reported an improvement of mean somatisation score from 1.57/5 before surgery to 1.23/5 a year after, an improvement of 0.34/5 ($p = 0.001$). Since symptoms were below the level of "mild" before treatment, the improvement may be of little value.

Self esteem
Lomholt et al 2016 reported a rise in patients' mean self-esteem scores from 83.0/100 before surgery to 89.3/100 six months after ($p<0.0001$). Parents also rated their child's self-esteem as improved, from 77.0/100 to 83.7/100 over the same period.

Behaviour
Lomholt et al 2016 reported that patients' mean behaviour scores improved from 87.4/100 before surgery to 89.9/100 six months after surgery ($p=0.05$). Parents' mean scores for their child's behaviour did not improve significantly.

Family activities and family cohesion
Lomholt et al 2016 and Jacobsen et al 2010 reported family activities and family cohesion scores.

Lomholt et al 2016 reported that patients' mean family activity scores improved from 88.0/100 before surgery to 95.2/100 six months after surgery ($p=0.001$). Parents' mean scores for family cohesion also improved from 89.0/100 to 95.2/100 over the same period ($p<0.0001$). Patients reported no change in family cohesion, but parents' mean scores improved from 79.1/100 to 84.8/100 over the six months ($p=0.03$).

Jacobsen et al 2010 reported better family activities scores in patients who had had surgery for pectus deformity (90.6/100) than in age-matched healthy controls (82.6/100) ($p<0.001$).

In patients who undergo surgery, is there any relationship between the degree of the pectus deformity (as quantified by the Haller index or other objective assessment) and the change in psychological outcomes?

No evidence relevant to this question was found.

Are there any sub-groups in which surgery produces a greater improvement in mental wellbeing than others?

No evidence relevant to this question was found.

How does the incorporation of psychosocial therapies into surgical management affect outcome?

No evidence relevant to this question was found.

5 Discussion

There are a number of serious weaknesses in the evidence that was found.

The studies were not well-controlled. Four of the studies (Luo et al 2017, Lomholt et al 2016, Kuru et al 2015 and Kelly et al 2008) used a before-and-after design with no patient control group, which makes it impossible to discern whether any changes observed are because of the intervention or because of another factor, such as the passage of time or some other change in participants' circumstances. Bahadir et al 2017's use of unoperated patients with higher pre-operative body satisfaction as controls is clearly inappropriate, because the reported differences are likely to reflect disease severity rather than the effect of surgery. Jacobsen et al 2011's use of healthy controls provides no information on the difference that surgery may make to those with pectus deformity. This study reported better results in patients after surgery than in healthy controls, which the authors said was "unexpected". Jacobsen et al 2010 suggest that greater parental attention and care to operated patients compared to controls may explain these findings. If so, the results shed no light on the procedure's clinical effectiveness, and indicate the biases which follow the use of unsuitable controls.

Although the studies all reported statistically significant results, *they provide little information on the significance to patients of the differences that they found*. A change may be large enough to make chance an unlikely explanation, but too small to be of real value to the patient. In many cases, patients' and parents' scores did not indicate the existence of major psychosocial or other difficulties before surgery, limiting the improvement that surgery could provide. Kelly et al 2008, for example, state that "All standardized effect sizes were very large (Cohen's d values ranged from 1.02 to 1.75)". However, Cohen's d is a measure of how far the means of the two groups are apart, as a proportion of the standard deviation; it provides no help in understanding whether the change in the measure under consideration is substantial enough to be meaningful to patients.

The outcome measures varied between studies and were often opaque. The terminology used in the technical reporting of psychometric questionnaires is often not suitable for publications read by surgeons and commissioners. Because the authors did not make clear what each outcome measure meant and what effect a change might have in a patient's life, it is difficult to assess the relevance and importance of the differences reported. The studies were remarkably free of any objective measures, such as school attendance or participation in sport.

There were many *important data the authors did not report*, such as whether participants had undergone other treatment before or after surgery, the extent to which activities of daily living were prevented by their deformity and how often the procedure had adverse effects.

The *involvement of parents* in the studies raises two issues. Four studies (Lomholt et al 2016, Kuru et al 2015, Kelly et al 2008, and Bahadir et al 2017) reported questionnaire results from parents as well as patients. First, the authors do not explore the validity of parents' assessments of their child's mental health, psycho-social functioning and self-esteem. Adolescents in particular may not share all aspects of their lives with parents, and are often not under parental observation. Second, parents may have influenced patients' questionnaire responses, especially in the case of younger children. Both these factors may have introduced bias into the studies' results.

These issues make it very difficult to draw conclusions from the studies that were found. Better reported studies with more appropriate controls are needed before one can be confident about the effect of surgery for pectus deformities on psychological and related outcomes.

6 Conclusion

The studies that were found report responses to questionnaires completed by patients and their parents. These suggest that some improvements in various psychological, social and behavioural measures may follow repair of pectus deformities. These include social functioning, emotional and behavioural problems, mental health, body image and somatisation, self-esteem, behaviour and family activities and family cohesion.

However, there are serious weaknesses in the studies. They were not well-controlled, and the significance to patients of the reported differences was unclear. In many cases, patients' and parents' scores did not indicate the existence of major psychosocial or other difficulties before surgery, limiting the improvement that surgery could provide. The outcome measures varied between studies and were often opaque.

Because of this, it is unclear the extent to which the changes are attributable to surgery and whether they are large enough to be of value.

No evidence was found about the relationship between the degree of the pectus deformity and change in psychological outcomes, about sub-groups in which surgery produces a greater improvement in mental wellbeing and whether the incorporation of psychosocial therapies into surgical management affects outcome.

The evidence that was found is not sufficient to conclude that the psychological, social and behavioural benefits of surgical treatment of pectus deformities are sufficient to justify its use.

7 Evidence Summary Tables

Surgical correction for pectus deformity – before and after surgery comparison									
Study reference	**Study design**	**Population characteristics**	**Intervention**	**Outcome measure type**	**Outcome measures**	**Results**	**Quality of evidence score**	**Applicability**	**Critical appraisal summary**
Luo L et al 2017	P1: Prospective uncontrolled cohort study (before and after design) 1 centre, 1 surgeon, China, 2009-2012	266 patients (aged ≥11, mean 19.02 ± SD[5] 4.42) with PE admitted for surgery. Patients excluded if they have: • had previous surgery • a major psychiatric disorder • recurrent PE • IQ below 70 • other complex chest wall deformity	MIRPE surgery	Primary outcome Clinical effectiveness	Somatisation (Chinese version of the Symptom Checklist-90 (SCL-90))[6]	Results reported as mean scores ± SD: Preoperative (7 days before surgery: 1.57 ± 0.59. Postoperative (1 year after surgery) 1.23 ± 0.62. Improvement of 0.34, p = 0.001	5	Direct	The authors provide no information on the validation of the questionnaires they used, including in the younger age group whom they studied (33% of participants were 11-16 years of age). They do not report maximum or minimum scores, or define what change in score is clinically meaningful. The method of questionnaire administration and any other possible confounding factors were not described, and the definition of mental health problems was imprecise. Thirteen tests were used without a Bonferroni correction. The corrected p-value for significance is 0.05/13 = 0.0038. Three of the results attained this level of significance.
				Primary outcome Clinical effectiveness	Interpersonal sensitivity (Chinese version of the Symptom Checklist-90 (SCL-90))	Preoperative 1.92 ± 0.72. Postoperative 1.77 ± 0.64. Improvement of 0.15, p = 0.025 (not significant after Bonferroni adjustment).			
				Primary outcome Clinical effectiveness	Anxiety (Chinese version of the Symptom Checklist-90 (SCL-90))	Preoperative 1.73 ± 0.71. Postoperative 1.58 ± 0.73. Improvement of 0.15, p = 0.013 (not significant after Bonferroni adjustment).			
				Primary outcome Clinical effectiveness	Mental health problems (Chinese version of the Symptom Checklist-90 (SCL-90))	Patients above threshold for diagnosis of mental health problems: preoperative 161/266 (60.5%), postoperative 79/266 (29.7%), improvement of 30.8%, p < 0.001.			
				Primary	Depressive Status	Proportion of participants above threshold for diagnosis of			

[5] Standard deviation
[6] The SCL-90 is a psychiatric self-report inventory containing 90 question chapters on 10 items, each scored 1 to 5 with higher scores indicating worse symptoms. A total score above 160, number of positive items above 43 or a 'certain factor' above 2, indicates that a patient was positive for mental health problems. Patients scoring above 160 on the SCL-90 or having scores of 3.0 or more for depression or anxiety items were asked to complete the Self-Rating Depression Scale (SDS) survey.

				outcome Clinical effectiveness	(Self-rating Depression Scale[7])	depression: preoperative 153/266 (57.5%), postoperative 76/266 (28.6%), improvement of 28.9%, p < 0.001.			
Lomholt JJ, et al 2016	P1:Prospective controlled cohort study (before and after design plus healthy control group) 1 centre, 1 surgeon, Denmark, 2006-2008	107 patients (aged 11-20) with PE and 106 of their parents People excluded if: pre-existing chronic diseases and cognitive defects present Healthy controls (n=183), age and gender-matched with patient group	MIRPE surgery for patients	Primary outcome Clinical effectiveness	Role/social: emotional Child health questionnaire (CHQ-CF87)[8]	Patients' mean scores: pre-surgery 90.6 (SD=15.8); 3 months post-surgery 96.5 (11.9), improvement of 5.9, p=0.002; 6 months post-surgery 98.7 (5.0), improvement of 8.1, p<0.0001.	7	Direct	Patients and their parents completed CHQ-CF87 and CHQ-PF50 questionnaires before surgery and at 3 and 6 months after surgery. The authors also report a comparison of pre-surgical scores with those of healthy controls, but this is out of the scope of the PICO. Of the 106 patients and one of their parents completing a baseline assessment before surgery, 85 (80%) completed the 3 and 6 month follow-up. There was no comparison of this group and their surgical outcomes with the group that did not complete the follow-up questionnaires. The CHQ tool is validated for the assessment of general health-related quality-of-life and is not specific to pectus deformity. The authors do not report maximum or minimum scores, or define what change in score is clinically meaningful, though they describe the effect size, in statistical terms, as "moderate-to-large". Parents' scores may be less valid because of lack of first-hand knowledge of the benefits and adverse effects of surgery; their scores tended to be higher than those of patients, though there is no statistical testing of this. The correlation between patients' and parents' scores, though statistically significant, was not strong (r=0.27 to 0.54, p < 0.0001). Due to the large number of statistical tests, a Bonferroni correction for multiple comparisons was carried out.
				Primary outcome Clinical effectiveness	Role/social: behavioural Child health questionnaire (CHQ-CF87)	Patients' mean scores: pre-surgery 94.8 (SD=12.0); 3 months post-surgery 97.6 (9.2), improvement of 2.8, p=0.20; 6 months post-surgery 99.2 (4.6), improvement of 4.4, p=0.004.			
				Primary outcome Clinical effectiveness	Role/social: emotional and behavioural combined Child health questionnaire (CHQ-PF50)[9]	Parents' mean scores: pre-surgery 89.6 (SD=20.0); 3 months post-surgery 94.8 (13.2) improvement of 5.2, p=0.06; 6 months post-surgery 98.5 (6.6), improvement of 8.9, p=0.001.			
				Primary outcome Clinical effectiveness	Behaviour Child health questionnaire (CHQ-CF87) for patients Child health questionnaire (CHQ-PF50) for parents of patients.	Patients' mean scores: pre-surgery 87.4 (SD=8.3); 3 months post-surgery 88.5 (7.3), improvement of 1.1, p=0.66; 6 months post-surgery 89.9 (7.8), improvement of 2.5, p=0.05. Parents' mean scores: pre-surgery 85.8 (SD=9.8); 3 months post-surgery 87.3 (10.1), improvement of 1.5, p=0.28; 6 months post-surgery 87.4 (9.6), improvement of 1.6, p=0.30.			
				Primary outcome Clinical effectiveness	Mental health Child health questionnaire (CHQ-CF87) for patients	Patients' mean scores: pre-surgery 82.6 (SD=11.1); 3 months post-surgery 85.7 (11.2), improvement of 3.1, p=0.07; 6 months post-surgery 86.2 (9.0), improvement of 3.6,			

[7] The Self-rated Depression Scale (SDS) is a measure depressive status. It consists of a 20-item questionnaire with responses ranging from 1 to 4, higher scores indicating more frequent depressive symptoms. A score of 53 or more indicates depression.

[8] The child self-reported version of the CHQ (CHQ-CF87) was developed for completion by children from ages 10 and older and consists of 10 multi-item scales and 4 single-item scales and. Scores for each subscale and single items are transformed on a scale from 0 (worst health) to 100 (best health) (except for the 'Change in Health' scale which ranges from 1 to 5).

[9] The long parent-report questionnaire (CHQ-PF50) consists of 11 multi-item scales and 4 single-item scales. As with the CHQ-CF87, scores for each subscale and single items are transformed on a scale from 0 (worst health) to 100 (best health) (except for the 'Change in Health' scale which ranges from 1 to 5).

					Child health questionnaire (CHQ-PF50) for parents of patients.	p=0.04. Parents' mean scores: pre-surgery 84.9 (SD=9.8); 3 months post-surgery 87.6 (11.8), improvement of 2.7, p=0.14; 6 months post-surgery 87.9 (9.2), improvement of 3.0, p=0.04.			
				Primary outcome Clinical effectiveness	Self-esteem Child health questionnaire (CHQ-CF87) for patients Child health questionnaire (CHQ-PF50) for parents of patients	Patients' mean scores: pre-surgery 83.0 (SD=12.0); 3 months post-surgery 87.1 (11.0), improvement of 4.1, p=0.004; 6 months post-surgery 89.3 (11.3), improvement of 6.3, p<0.0001. Parents' mean' scores: pre-surgery 77.0 (SD=14.1); 3 months post-surgery 80.7 (14.6), improvement of 3.7, p=0.05; 6 months post-surgery 83.7 (16.0), improvement of 6.7, p=0.003.			
				Primary outcome Clinical effectiveness	Family activities Child health questionnaire (CHQ-CF87) for patients Child health questionnaire (CHQ-PF50) for parents of patients	Patients' mean scores: pre-surgery 88.0 (SD=14.2); 3 months post-surgery 91.5 (10.7), improvement of 3.5, p=0.13; 6 months post-surgery 95.2 (9.9), improvement of 7.2, p=0.001. Parents' mean scores: pre-surgery 89.0 (SD=11.9); 3 months post-surgery 95.1 (7.4), improvement of 6.1, p<0.0001; 6 months post-surgery 95.2 (8.7), improvement of 6.2, p<0.0001.			
				Primary outcome Clinical effectiveness	Family cohesion Child health questionnaire (CHQ-CF87) for patients Child health questionnaire (CHQ-PF50) for parents of patients.	Patients' mean scores: pre-surgery 79.7 (SD=21.8); 3 months post-surgery 81.4 (18.6), improvement of 1.7, p=1.00; 6 months post-surgery 81.5 (20.2), improvement of 1.8, p=1.00. Parents' mean scores: pre-surgery 79.1 (SD=18.4); 3 months post-surgery 82.0 (15.0), an improvement of 2.9, p=0.33; 6 months post-surgery 84.8 (12.9), improvement of 5.7, p=0.03.			
Kuru P, et al 2015	P1:Prospective uncontrol	88 patients (aged 14-29) with PE	Patients underwent MIRPE	Primary outcome	Psychosocial functioning measured via	Patients' scores (median, inter-quartile range): pre-surgery 22.5 (19 to 25), 6 months post-surgery 33 (30	7	Direct	The authors claim that the Pectus Excavatum Evaluation Questionnaire, on which the Nuss questionnaire is based, has been validated, but cite

	led cohort study (before and after design) 1 centre, Turkey, number of surgeons and dates not reported	and their parents (n for parents not provided)	surgery	Clinical effectiveness	the Nuss questionnaire modified for adults[10]	to 35), improvement of 10.5 (6 to 13), p=0.000. Parents' scores (median, inter-quartile range): pre-surgery 20 (17 to 23), 6 months post-surgery 24 (21 to 26), improvement of 4 (1 to 7), p=0.000.			in support of this a study which reported only 22 parents and 19 children (Lawson et al 2003), casting doubt on the adequacy of the validation. The authors do not define what change in score is clinically meaningful. The method of questionnaire administration and any other possible confounding factors were not described. Parents' scores may be less valid because of lack of first-hand knowledge of the benefits and adverse effects of surgery.
Kelly R. et al 2008	P1 Prospective uncontrolled cohort study (before and after design) Multi-centre (11 hospitals), US, 2001-2006	264 patients (aged 8-21) with PE and 291 of their parents.	Patients underwent MIRPE or Ravitch surgery	Primary outcome Clinical effectiveness	Body image scores measured via the Pectus Excavatum Evaluation Questionnaire (PEEQ)[9]	247 patients (87%) completed both pre- and post-surgery questionnaires. Patients' mean scores: pre-surgery 2.30 (SD 0.62); 1 year post-surgery 1.40 (0.42); standardised effect size 1.70. p<0.0001.	6	Direct	The validation study for the Pectus Excavatum Evaluation Questionnaire, used by Kelly et al 2008, reported only 22 parents and 19 children (Lawson et al 2003), casting doubt on the adequacy of the validation. The authors do not report maximum or minimum scores, or define what change in score is clinically meaningful. Parents' scores may be less valid because of lack of first-hand knowledge of the benefits and adverse effects of surgery.
				Primary outcome Clinical effectiveness	Emotional difficulties scores measured via Pectus Excavatum Evaluation Questionnaire (PEEQ)[9]	274 (94%) of parents completed both pre- and post-surgery questionnaires. Parents' mean scores: pre-surgery 1.81 (SD 0.70); 1 year post-surgery 1.24 (0.36); standardised effect size 1.02, p<0.0001.			
				Primary outcome Clinical effectiveness	Social self-consciousness scores measured via Pectus Excavatum Evaluation Questionnaire (PEEQ)[9]	Parents' mean score: pre-surgery 2.86 (SD 1.03); 1 year post-surgery 1.33 (0.68); standardised effect size 1.75, p<0.0001.			

[10] The Pectus Excavatum Evaluation Questionnaire (PEEQ) is a disease-specific quality-of-life instrument for children, later modified for adults and termed the Nuss Questionnaire or NQ-mA. The patient version has 12 items (possible score range 12 to 48) and the parent version has 11 (possible score range 11 to 44). Higher scores indicate a better quality of life.

Surgical correction for pectus deformity – comparison of surgery with no surgery for PE or PC									
Study reference	**Study Design**	**Population characteristics**	**Intervention**	**Outcome measure type**	**Outcome measures**	**Results**	**Quality of Evidence Score**	**Applicability**	**Critical Appraisal Summary**
Bahadir AT, et al 2017	P1 prospective controlled study 1 centre, Turkey, number of surgeons and dates not reported	63 patients (aged 9-17) with pectus deformity and 63 of their parents	32 patients (75% PE, 25% PC) had MIRPE or MIRPC surgery (at least 6 months before study entry) 31 patients (47% PE, 54% PC) did not have surgery	Primary outcomes Clinical effectiveness	Psychiatric rating scales: State-Trait Anxiety Inventory for Children[11]	Mean scores (SD): operated: 32.28 (5.83); non-operated: 34.9 (6.73), difference -2.62, p=0.201.	5	Direct	This unrandomised comparison of operated and non-operated patients is at risk of serious confounding. The authors do not report why participants did or did not receive surgery, beyond noting that "patients in the operated group mostly decided to come to the hospital themselves". The authors do not report on the severity of anatomical abnormality in the two groups, but report that a higher proportion of patients receiving an operation had a lower pre-operative satisfaction with body image score (3.4/10 versus 5.0/10, p=0.019). Those who successfully sought surgery are likely to have been more severely affected by their condition, so any reported psychological differences may reflect the impact of disease severity rather than that of treatment. There are different proportions of children with PE and PC in the control and intervention group (PE = 75% in the intervention group and 46.7% in the control group). The authors claim that one result (prosocial problems in the SDQ-SR version) showed a statistically significant difference between the two groups. However, the authors carried out 16 tests of statistical significance and did not use a Bonferroni correction. The corrected p-value for significance is 0.05/16 = 0.0032. None of the results attained this level of significance, making it necessary to conclude that the single apparently significant result was due to chance.
				Primary outcomes Clinical effectiveness	Psychiatric rating scales: Child Depression Inventory[12]	Mean scores (SD): operated: 11.13 (5.82); non-operated: 10.61 (8.01), differences 0.52, p=0.311.			
				Primary outcomes Clinical effectiveness	Psychiatric rating scales: SDQ-PR[13] Version	The authors reported emotional symptoms, conduct problems, hyperactivity/inactivity, peer relationship problems and pro-social behaviour. None showed a statistically significant difference after Bonferroni correction.			
				Primary outcomes Clinical effectiveness	Psychiatric rating scales: SDQ-SR Version	The authors reported total score, emotional problems, conduct problems, hyperactivity/inactivity, peer relationship problems and prosocial behaviour. None showed a statistically significant difference after Bonferroni correction.			

[11] The State-Trait Anxiety Inventory for Children – Trait Version (STAIC-T) has two subscales, each containing 20 items that assesses state and trait anxiety. The STAIC-T measures trait anxiety on a 3-point Likert scale. Higher scores mean higher levels of anxiety.

[12] The Children's Depression Inventory (CDI and CDI2) is a psychological assessment that rates the severity of symptoms related to depression or dysthymic disorder in children and adolescents. The CDI is a 27-item scale that is self-rated and symptom-oriented. Higher scores indicate increasing severity of depression.

[13] The Strengths and Difficulties Questionnaire (SDQ) is a 25-item behavioural screening questionnaire for children and adolescents. There is a self-reported version (SDQ-SR) and a parent reported version (SDQ-PR). The SDQ has 5 subscales and 5 items per subscale scored on a 3-point Likert scale. Higher scores indicate more emotional and behavioural problems.

Surgical correction for pectus deformity – comparison after surgery with healthy controls									
Study reference	**Study Design**	**Population characteristics**	**Intervention**	**Outcome measure type**	**Outcome measures**	**Results**	**Quality of Evidence Score**	**Applicability**	**Critical Appraisal Summary**
Jacobsen EB et al 2010	P1 Prospective controlled study 1 centre, Denmark, 2003-2005, number of surgeons not reported	119 children and adolescents (aged 8 to 20) receiving surgery for PE , and their parents (n not reported) 271 healthy control children (aged 9 to 20 years) and 147 of their parents also completed questionnaires	Patients had surgery for PE, procedure not reported	Primary outcome Clinical effectiveness	CHQ87[14]: role function: emotional	Mean scores (SD): operated 96.7 (5.83); controls: 90.6 (16.8), difference 6.1, p<0.001.	6	Direct	No parental survey results relevant to the PICO were reported. The control group were matched by age but not gender (girls were 52% in control group and 14% in intervention group) or other variables which could introduce bias to the overall results. Questionnaire sent out between 6 and 30 months after surgery. No baseline assessment of this before surgery. The authors carried out 42 tests of statistical significance but did not use a Bonferroni correction. The corrected p-value for significance is 0.05/42 = 0.00119. Results reported here are only those which attained this level of significance and were within scope of this review. Jacobsen et al 2010 suggest that greater parental attention and care to operated patients compared to controls may explain their "unexpected" findings. If so, the results shed no light on the procedure's clinical effectiveness. Other issues which may compromise the internal validity of the results include: the varying time between surgery and completing the questionnaires for the intervention group (6 to 30 months) and the potential for direct or indirect involvement of parents in questionnaire responses given by patients.
				Primary outcome Clinical effectiveness	CHQ87: role function: behavioural	Mean scores (SD): operated 98.2 (5.83); controls: 95.4 (16.8), difference 2.8, p<0.001.			
				Primary outcome Clinical effectiveness	CHQ87: mental health	Mean scores (SD): operated 83.7 (12.5); controls: 78.4 (14.8), difference 5.3, p<0.001.			
				Primary outcome Clinical effectiveness	CHQ87: family activities	Mean scores (SD): operated 90.6 (15.8); controls: 82.6 (18.1), difference 8.0, p<0.001.			

[14] The child self-reported version of the CHQ (CHQ-CF87) was developed for completion by children from ages 10 and older and consists of 10 multi-item scales and 4 single-item scales. Scores for each subscale and single items are transformed on a scale from 0 (worst health) to 100 (best health) (except for the 'Change in Health' scale which ranges from 1 to 5)..

8 Grade of Evidence Table

Surgical correction for pectus deformity – before and after surgery comparison					
Outcome Measure	**Reference**	**Quality of Evidence Score**	**Applicability**	**Grade of Evidence**	**Interpretation of Evidence**
Somatisation	Luo et al 2017	**5**	**Direct**	**C**	Somatisation is not defined in the papers included in this rapid evidence review, but usually means the manifestation of psychological distress by the presentation of bodily symptoms. Luo et al 2017 report a pre-surgery (7 days before surgery) mean score of 1.57 (out of a total score of 5, where 1 equals 'no' symptoms and 2 equals 'mild' symptoms) and a post-surgery (1 year after surgery) mean score of 1.23, an improvement of 0.34, p = 0.001. This suggests that the procedure may reduce somatisation, but the low reliability of Luo et al 2017 casts doubt on this. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Since symptoms were below "mild" before treatment, the improvement may be of little value. Reduced somatisation would be of benefit to patients, but Luo et al 2017 does not provide a secure basis for conclusions about this outcome.
Interpersonal sensitivity	Luo et al 2017	**5**	**Direct**	**C**	Interpersonal sensitivity is not defined in the papers included in this rapid evidence review, but usually means the ability to read other people's feelings and states, and to respond appropriately. Luo et al 2017 report no significant change in patients' interpersonal sensitivity, after Bonferroni correction for the use of multiple tests. This suggests that the procedure does not affect interpersonal sensitivity. Improved interpersonal sensitivity would be of benefit to patients, but Luo et al 2017 does not indicate that the procedure improves it.
Role/social limitations: emotional, emotional difficulties	Lomholt et al 2016 Kelly et al 2008	**7** **6**	**Direct** **Direct**	**B**	Role/social limitation: emotional is defined in Lomholt et al 2016 as a limitation in school work/play with friends due to sadness/worry in the last four weeks. Lomholt et al 2016 report patients' mean scores pre-surgery of 90.6 (out of a total score of 100, where 0 equals worst health and 100 equals best health), 3 months post-surgery of 96.5, an improvement of 5.9, p=0.002, and 6 months post-surgery of 98.7, an improvement of 8.1, p<0.0001. This suggests that the procedure may improve emotional health. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Improved emotional health would be of benefit to patients, but Lomholt et al 2016 does not provide a secure basis for conclusions about this outcome.

Anxiety	Luo et al 2017	**5**	**Direct**	**C**	Anxiety is not defined in the papers included in this rapid evidence review, but usually means a feeling of worry, nervousness or unease. Luo et al 2017 report a pre-surgery (7 days before surgery) mean score of 1.73 (out of a total score of 5, where 1 equals 'no' symptoms and 2 equals 'mild' symptoms) and a post-surgery (1 year after surgery) mean score of 1.58, an improvement of 0.15, p = 0.025. This suggests that the procedure may reduce anxiety, but the low reliability of Luo et al 2017 casts doubt on this. The result was not statistically significant after adjustment for the multiple tests reported by Luo et al 2017. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Reduced anxiety would be of benefit to patients, but Luo et al 2017 does not provide a secure basis for conclusions about this outcome.
Mental health problems	Luo et al 2017	**5**	**Direct**	**B**	Mental health problems are defined in Lomholt et al 2016 as amount of time feeling unhappy, lonely, nervous and worried in the last four weeks. Lomholt et al 2016 report patients' mean scores pre-surgery of 82.6, (out of a total score of 100, where 0 equals worst health and 100 equals best health), 3 months post-surgery of 85.7, an improvement of 3.1, p=0.07 and 6 months post-surgery of 86.2, improvement of 3.6, p=0.04. Lomholt et al 2016 report parents' mean scores pre-surgery of 84.9, 3 months post-surgery of 87.6, an improvement of 2.7, p=0.14 and 6 months post-surgery of 87.9, an improvement of 3.0, p=0.04. This suggests that the procedure may improve mental health problems. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Improved mental health problems would be of benefit to patients, but Lomholt et al 2016 does not provide a secure basis for conclusions about this outcome.
	Lomholt et al 2016	**7**	**Direct**		
Depression	Luo et al 2017	**5**	**Direct**	**C**	Depression is not defined in the papers included in this rapid evidence review, but usually means feelings of severe despondency and dejection. Luo et al 2017 report that the proportions of participants above a threshold for diagnosis of depression were preoperative 153/266 (57.5%), postoperative 76/266 (28.6%), an improvement of 28.9%, p < 0.001. This suggests that the procedure may reduce the prevalence of depression, but the low reliability of Luo et al 2017 casts doubt on this. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Reduced prevalence of depression would be of benefit to patients, but Luo et al 2017 does not provide a secure basis for conclusions about this outcome.
Role/social limitations: behavioural	Lomholt et al 2016	**7**	**Direct**	**B**	Role/social limitation: behavioural is defined in Lomholt et al 2016 as a limitation in school work/play with friends due limits in behaviour in the last four weeks.

					Lomholt et al 2016 report patients' mean scores pre-surgery of 94.8 (out of a total score of 100, where 0 equals worst health and 100 equals best health), 3 months post-surgery of 97.6, an improvement of 2.8, p=0.2 and 6 months post-surgery of 99.2, an improvement of 4.2, p=0.004. This suggests that the procedure may improve role/social: behavioural. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Improved role/social: behavioural might be of benefit to patients, but Lomholt et al 2016 does not provide a secure basis for conclusions about this outcome.
Role/social: emotional and behavioural combined parental score	Lomholt et al 2016	**7**	**Direct**	**B**	Role/social limitation: emotional is defined in Lomholt et al 2016 as a limitation in school work/play with friends due to sadness/worry in the last four weeks. Role/social limitation: behavioural is defined in Lomholt et al 2016 as a limitation in school work/play with friends due limits in behaviour in the last four weeks. Lomholt et al 2016 report parents' mean scores pre-surgery of 89.6 (out of a total score of 100, where 0 equals worst health and 100 equals best health), 3 months post-surgery of 94.8, an improvement of 5.2, p=0.06 and 6 months post-surgery of 98.5, an improvement of 8.9, p=0.001. This suggests that the procedure may improve role/social: emotional and behavioural might be of benefit to patients. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Improved role/social: emotional and behavioural function might be of benefit to patients, but Lomholt et al 2016 does not provide a secure basis for conclusions about this outcome.
Behaviour	Lomholt et al 2016	**7**	**Direct**	**B**	Behaviour is defined in Lomholt et al 2016 as the extent of bad behaviour compared to other children of the same age in the last four weeks. Lomholt et al 2016 report patients' mean scores pre-surgery of 87.4 (out of a total score of 100, where 0 equals worst health and 100 equals best health), 3 months post-surgery of 88.5, an improvement of 1.1, p=0.66 and 6 months post-surgery of 89.9, an improvement of 2.5, p=0.05. Lomholt et al 2016 report parents' mean scores pre-surgery of 85.8 (out of a total score of 100, where 0 equals worst health and 100 equals best health), 3 months post-surgery of 87.3, an improvement of 1.5, p=0.28 and 6 months post-surgery of 87.4, an improvement of 1.6, p=0.30. This suggests that the procedure may improve behaviour. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Improved behaviour would be of benefit to patients, but Lomholt et al 2016 does not provide a secure basis for conclusions about this outcome.
Self-esteem	Lomholt et al 2016	**7**	**Direct**	**B**	Self-esteem is defined in Lomholt et al 2016 as satisfaction with appearance, activities and interaction with friends/family in the last four weeks. Lomholt et al 2016 report patients' mean scores pre-surgery of 83.0 (out of a total score

					of 100, where 0 equals worst health and 100 equals best health), 3 months post-surgery of 87.1, an improvement of 4.1, p=0.004 and 6 months post-surgery of 89.3, an improvement of 6.3, p<0.001. Lomholt et al 2016 report parents' mean scores pre-surgery of 77.0 (out of a total score of 100, where 0 equals worst health and 100 equals best health), 3 months post-surgery of 80.7, an improvement of 3.7, p=0.0 and 6 months post-surgery of 83.7, an improvement of 6.7, p=0.003. This suggests that the procedure may improve self-esteem. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Improved self-esteem would be of benefit to patients, but Lomholt et al 2016 does not provide a secure basis for conclusions about this outcome.
Family activities	Lomholt et al 2016	**7**	**Direct**	**B**	Family activities are defined in Lomholt et al 2016 as limitations in family activities due to behaviour/health in the last four weeks. Lomholt et al 2016 report patients' mean scores pre-surgery of 88.0 (out of a total score of 100, where 0 equals worst health and 100 equals best health), 3 months post-surgery of 91.5, an improvement of 3.5, p=0.13 and 6 months post-surgery of 95.2, an improvement of 7.2, p=0.001. Lomholt et al 2016 report parents' mean scores pre-surgery of 89.0 (out of a total score of 100, where 0 equals worst health and 100 equals best health), 3 months post-surgery of 95.1, an improvement of 6.1, p<0.001 and 6 months post-surgery of 95.2, an improvement of 6.2, p<0.001. This suggests that the procedure may improve family activities. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Improved family activities would be of benefit to patients, but Lomholt et al 2016 does not provide a secure basis for conclusions about this outcome.
Family cohesion	Lomholt et al 2016	**8**	**Direct**	**B**	Family cohesion is defined in Lomholt et al 2016 as the family's ability to get along with one another. Lomholt et al 2016 report patients' mean scores pre-surgery of 79.7 (out of a total score of 100, where 0 equals worst health and 100 equals best health), 3 months post-surgery of 81.4, an improvement of 1.7, p=1.00 and 6 months post-surgery of 81.5, an improvement of 1.8, p=1.00. Lomholt et al 2016 report parents' mean scores pre-surgery of 79.1 (out of a total score of 100, where 0 equals worst health and 100 equals best health), 3 months post-surgery of 82.0, an improvement of 2.9, p=0.33 and 6 months post-surgery of 84.8, an improvement of 5.7, p=0.03. This suggests that the procedure may improve family cohesion from the perspective of parents but not patients. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective.

					Improved family cohesion would be of benefit to patients, but Lomholt et al 2016 does not provide a secure basis for conclusions about this outcome.
Psychosocial functioning	Kuru et al 2015	**7**	**Direct**	**B**	Psychosocial functioning is not defined in the papers included in this rapid evidence review, but usually means the interrelation of social factors and individual thought and behaviour. Kuru et al 2015 report patients' mean scores pre-surgery of 22.5 (out of a total score of 48, where higher scores mean better health status) and 6 months post-surgery of 33, an improvement of 10.5, p=0.00. Kuru et al 2015 report parents' median scores pre-surgery of 20 (out of a total score of 44, where higher scores mean better health status) and 6 months post-surgery of 24, an improvement of 4, p=0.00. This suggests that the procedure may improve psychosocial functioning. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Improved psychosocial functioning would be of benefit to patients, but Kuru et al 2015 does not provide a secure basis for conclusions about this outcome.
Body Image	Kelly et al 2008	**6**	**Direct**	**C**	Body image is not defined in the papers included in this rapid evidence review, but usually means a person's perception of the attractiveness of their own body. Kelly et al 2008 report patients' median scores pre-surgery of 2.3 (out of a total score of 4, where lower scores mean better health status and 1 = very happy) and 1 year post-surgery of 1.4, a standardised effect size of 1.70. p<0.0001. This suggests that the procedure may improve body image. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Improved body image would be of benefit to patients, but Kelly et al 2008 does not provide a secure basis for conclusions about this outcome.
Emotional difficulties	Kelly et al 2008	**6**	**Direct**	**C**	Emotional difficulties are not defined in the papers included in this rapid evidence review. Kelly et al 2008 report parents' mean scores pre-surgery of 1.81 (out of a total score of 4, where lower scores mean better health status and 1 = very happy) and 6 months post-surgery of 1.24, a standardised effect size of 1.02, p<0.0001. This suggests that the procedure may improve emotional difficulties from parents' perspectives. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Reduced emotional difficulties would be of benefit to patients, but Kelly et al 2008 does not provide a secure basis for conclusions about this outcome.
Social self-consciousness	Kelly et al 2008	**6**	**Direct**	**C**	Social self-consciousness is not defined in the papers included in this rapid evidence review, but usually means one's heightened sense of self-awareness or preoccupation with oneself. Kelly et al 2008 report parent's mean scores pre-surgery of 2.86 (out of a total score of 4, where lower scores mean better health status and 1 = very happy) and post-surgery of

					1.33, a standardised effect size of 1.75, p<0.0001. This suggests that the procedure may improve social self-consciousness from parents' perspectives. The clinical significance of a change of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Improved social self-consciousness would be of benefit to patients, but Kelly et al 2008 does not provide a secure basis for conclusions about this outcome.

Surgical correction for pectus deformity – comparison of surgery with no surgery					
Outcome Measure	**Reference**	**Quality of Evidence Score**	**Applicability**	**Grade of Evidence**	**Interpretation of Evidence**
Anxiety	Bahadir et al 2017	5	Direct	C	Anxiety is not defined in the papers included in this rapid evidence review, but usually means a feeling of worry, nervousness or unease. Bahadir et al 2017 report mean scores in operated patients of 32.28 (out of a total score of 120, where lower scores mean better health status) and in non-operated patients of 34.9, a difference of -2.62, p=0.201. This suggests that the procedure does not reduce anxiety. Reduced anxiety would be of benefit to patients, but Bahadir et al 2017 does not provide a basis for concluding that the procedure leads to this.
Depression	Bahadir et al 2017	5	Direct	C	Depression is not defined in the papers included in this rapid evidence review, but usually means feelings of severe despondency and dejection. Bahadir et al 2017 report mean scores in operated patients of 11.13 (out of a total score of 81, where lower scores mean better health status) and in non-operated patients of 10.61, a difference of 0.52, p=0.311. This suggests that the procedure does not reduce depression. Reduced depression would be of benefit to patients, but Bahadir et al 2017 does not provide a basis for concluding that the procedure leads to this.

Surgical correction for pectus deformity – comparison after surgery with healthy controls					
Outcome Measure	**Reference**	**Quality of Evidence Score**	**Applicability**	**Grade of Evidence**	**Interpretation of Evidence**
Role/social limitation: emotional	Jacobsen et al 2010	6	Direct	C	Role/social limitation: emotional is defined in Lomholt et al 2016 as a limitation in school work/play with friends due to sadness/worry in the last four weeks. Jacobsen et al 2010 report mean scores in operated patients of 96.7 (out of a total score of 100, where 0 equals worst health and 100 equals best health) and in healthy controls of 90.6, a difference of 6.1, p<0.001. This suggests that those who undergo the procedure may report better role function: emotional than healthy controls. The clinical significance of a difference of this size is not

					reported and unclear, making the result hard to interpret from a patient's perspective. Improved role function emotional might be of benefit to patients, but the design of Jacobsen et al 2010's study means that it provides no information about whether the procedure leads to this outcome.
Role/social limitation: behavioural	Jacobsen et al 2010	6	Direct	C	Role/social limitation: behavioural is defined in Lomholt et al 2016 as a limitation in school work/play with friends due limits in behaviour in the last four weeks. Jacobsen et al 2010 report mean scores in operated patients of 98.2 (out of a total score of 100, where higher scores mean better health status) and in healthy controls of 95.4, a difference of 2.8, p<0.001. This suggests that those who undergo the procedure may report better role function: behavioural than healthy controls. The clinical significance of a difference of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Improved role function behavioural might be of benefit to patients, but the design of Jacobsen et al 2010's study means that it provides no information about whether the procedure leads to this outcome.
Mental health problems	Jacobsen et al 2010	6	Direct	C	Mental health problems are defined in Lomholt et al 2016 as amount of time feeling unhappy, lonely, nervous and worried in the last four weeks. Jacobsen et al 2010 report mean scores in operated patients of 83.7 (out of a total score of 100, where 0 equals worst health and 100 equals best health) and in healthy controls of 78.4, a difference of 5.3, p<0.001. This suggests that those who undergo the procedure may report better mental health than healthy controls. The clinical significance of a difference of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Improved mental health would be of benefit to patients, but the design of Jacobsen et al 2010's study means that it provides no information about whether the procedure leads to this outcome.
Family activities	Jacobsen et al 2010	6	Direct	C	Family activities are defined in Lomholt et al 2016 as limitations in family activities due to behaviour/health in the last four weeks. Jacobsen et al 2010 report mean scores in operated patients of 90.6 (out of a total score of 100, where 0 equals worst health and 100 equals best health) and in healthy controls of 82.6, a difference of 8.0, p<0.001. This suggests that those who undergo the procedure may report better family activities than healthy controls. The clinical significance of a difference of this size is not reported and unclear, making the result hard to interpret from a patient's perspective. Improved family activities might be of benefit to patients, but the design of Jacobsen et al 2010's study means that it provides no information about whether the procedure leads to this outcome.

9 Literature Search Terms

Search Terms Indicate all terms to be used in the search	
P – Patients / Population Which patients or populations of patients are we interested in? How can they be best described? Are there subgroups that need to be considered?	All adolescent and adult patients with pectus excavatum or pectus carinatum. Supporting information: Pectus deformities are common and are present in 1 in 400 children, more commonly affecting boys. They affect the anterior chest wall with the vast majority of patients being affected to only a very minor degree. There are 2 basic types: pectus excavatum (PE), or "funnel chest", and pectus carinatum (PC), or "pigeon chest". PE is more common. 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. Pectus deformities significantly impair the physical, social and psychological wellbeing of the individuals affected. In severe deformity, particularly severe PE, cardiac and/or respiratory function may also be impaired. Many patients are seen for advice in thoracic surgical clinics, and do not require treatment. A small number are considered for various forms of surgery. For the year 2011-12, 311 pectus operations were carried out in the whole of the UK (SCTS Thoracic Surgery register). In addition to measures of cardiac and respiratory compromise, these assessments will include assessment of the degree of physical deformity (eg Haller index) and the level of psychological distress being experienced as described by PD specific measures together with broader QoL assessment tools
I – Intervention Which intervention, treatment or approach should be used?	Pectus surgery, with or without accompanying psychological therapy. Supporting information: There are two surgical procedures undertaken to correct the deformity, these are: Nuss (minimally invasive repair of pectus excavatum – MIRPE): This "keyhole surgery" procedure involves placing one or two curved steel bars inside the patient's chest behind the sternum, forcing it back into a more normal shape. The bars are left in place for several years and are then removed with a second operation. The Nuss procedure is generally only applicable in pectus excavatum. It involves small incisions at the side of the patient's chest and avoid the necessity for a scar at the front.

	Ravitch: This is an open operation suitable for both PE and PC. It is carried out through an incision at the front of the patient's chest. Cuts are made in the rib cartilages on each side and on the sternum to allow correction of the deformity. For PE, some form of support is usually required to support the sternum once it has been brought forward. This used to involve insertion of a metal bar (which would necessitate a second operation for removal) but nowadays a mesh is more likely to be used. Ravitch procedures can be used for any pectus abnormality including pectus carinatum and the more complex deformities.
C – Comparison What is/are the main alternative/s to compare with the intervention being considered?	No intervention Suction devices (vacuum) Bracing Implants Psycho-social therapies Breast augmentation Studies where multiple comparison interventions were considered (e.g. bracing and psycho-social therapies) should be included within this review.
O – Outcomes What is really important for the patient? Which outcomes should be considered? Examples include intermediate or short-term outcomes; mortality; morbidity and quality of life; treatment complications; adverse effects; rates of relapse; late morbidity and re-admission	The primary outcome for this review should be: Psychological outcome which may include but is not limited to measures of ; depression, anxiety, self-esteem, body image, social isolation and dysmorphic In addition to this secondary outcomes should include: Objectively assessed degree of improvement to appearance of the deformity and the relationship to the preoperative levels psychological distress Health Related Quality of life [For the purposes of this specific review, other clinical effectiveness outcomes will be out of scope of this evidence review and not reported. This is a departure from NHS England standard methodology]
Assumptions / limits applied to search Section to be completed in accordance with section 2.4	
Inclusion Criteria	All study types but not case series where n is less than 5 Peer reviewed journals only, English language publications in the last 10 yrs
Exclusion Criteria	

10 Search Strategy

The following databases were searched: Embase, PubMed, Cochrane Library and PsychINF, limiting the search to 10 years up to the search date of 9th November 2017. Conference abstracts, case reports, commentary, letters and editorials were excluded.

Embase search

# ▲	Searches	Results
1	funnel chest/su [Surgery]	1435
2	pigeon thorax/su [Surgery]	132
3	*funnel chest/	2055
4	*pigeon thorax/	282
5	((pectus or thorax or thoracic) adj2 (deformit* or abnormalit*)).ti,ab.	1363
6	(pectus excavatum or pectus excavatus or funnel chest or funnel thorax).ti,ab.	2652
7	(pectus carinatum or pectus carinatus or pigeon chest or pigeon thorax).ti,ab.	551
8	3 or 4 or 5 or 6 or 7	4309
9	thorax surgery/	31601
10	minimally invasive surgery/	33905
11	surgical technique/	317624
12	(surg* or repair* or operat* or procedur*).ti.	982281
13	(minimally invasive repair or mirpe or nuss).ti,ab.	930
14	ravitch.ti,ab.	263
15	9 or 10 or 11 or 12 or 13 or 14	1239510
16	8 and 15	1858
17	((pectus or thorax or thoracic) adj2 (deformit* or abnormalit*) adj5 (surg* or repair* or operat* or procedur*)).ti,ab.	180
18	((pectus excavatum or pectus excavatus or funnel chest or funnel thorax) adj5 (surg* or repair* or operat* or procedure*)).ti,ab.	1261
19	((pectus carinatum or pectus carinatus or pigeon chest or pigeon thorax) adj5 (surg* or repair* or operat* or procedur*)).ti,ab.	117
20	1 or 2 or 16 or 17 or 18 or 19	2273
21	psychological aspect/	475841
22	exp adaptive behavior/ or exp coping behavior/ or exp emotion/ or empowerment/	570163
23	"quality of life"/	372272
24	depression/ or mood disorder/	342060

25	anxiety disorder/ or anxiety/	221201
26	mental disease/	206291
27	self concept/ or body image/ or personal appearance/ or self esteem/	111173
28	emotional deprivation/ or social isolation/	20782
29	(psycho* or mental* or emotion* or feeling*).ti.	396731
30	((psycho* or mental or emotion*) adj2 (stress or adapt* or adjust*)).ti,ab.	38857
31	(depression or depressive or anxiety).ti,ab.	541211
32	(coping or empower* or body image or self-esteem or self image or dysmorph*).ti,ab.	132249
33	"quality of life".ti,ab.	335598
34	((social* adj2 (isolat* or exclu*)) or friend* or sociali*).ti,ab.	106532
35	21 or 22 or 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34	2186226
36	20 and 35	273
37	limit 36 to (english language and yr="2008 -Current")	162
38	conference*.pt.	3527350
39	37 not 38	110

11 Evidence Selection

Total number of publications reviewed: 26

Total number of publications considered potentially relevant: 18

Total number of publications selected for inclusion in this briefing: 6

12 References

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