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Review

Minimally invasive repair of pectus excavatum deformity

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Summary

This review is trying to address the effectiveness and sustainability of results following minimally invasive repair of pectus excavatum (MIRPE). The aim is to present these results for the benefit of clinicians and the patients. Literature search has revealed 179 hits, which were independently assessed and led to 80 publications being formally reviewed. Studies reporting results from less than 10 patients were excluded. Thirty-five studies were found to be reporting results from patients' and/or surgeons' perspective and they were included in this review. Data from the United Kingdom registry for MIRPE were also included. Results from over 2997 patients (age: <1—85 years) who had MIRPE and 1393 patients who had their metallic bar removed were assessed. The most common indication for surgery was cosmesis. There was a net gain with regard to self-esteem for 96—100% of the individuals. A percentage of procedures (0—20%) was assessed by surgeons as having an 'unsatisfactory outcome' and a number of patients (0—25%) reported an 'unsatisfactory end result.' However, these percentages are not necessarily referring to the same patients and an unsatisfactory result does not seem to affect the positive effect on self-esteem. The reported changes in social life, lung capacity, cardiovascular capacity, exercise capacity and general health are based on weak data and significant improvements, if any, are probably seen in a limited number of patients. The metallic bars were removed after 1.5—4.5 years and there is an overall 0—4.5% reported recurrence post-bar removal. In conclusion, MIRPE may improve cosmesis and self-esteem of patients with pectus excavatum deformity. Direct or indirect improvement in other physiological parameters may also help the 'well-being' of these patients and their social integration. There is a clear need for standardisation in the way results are reported in the literature and a socioeconomic analysis with regard to gains, benefits and costs related to MIRPE.

Keywords: MIRPE; Nuss procedure; Results; Outcome

1. Introduction

Pectus excavatum (PE) or 'funnel chest' is the most common deformity of the chest wall, accounting for 90% of all such deformities. Although the exact cause of this deformity remains unknown, up to 46% of patients have a family history. PE tends to affect males more often than females (9:1) with an overall incidence of 1 in 400—1000 births [1] in the United States.

The most commonly occurring problem in this group of patients is associated with the psychosocial impact that the deformity has on a developing personality and the patients' social integration. This is particularly true during adolescence when patients with PE tend to alter their behaviour to avoid activities requiring exposure of their chest. Patients with moderate-to-severe PE may also present with shortness of breath on exertion attributed to problems with thoracic wall compliance and cardiovascular compromise due to external compression and displacement of the heart [2]. Chest pain is also described by sufferers but it remains difficult to assess and treat as the cause remains uncertain.

There are a number of therapeutic options for the correction of PE deformity: plastic surgery with soft-tissue rearrangement (including fat injections and breast augmentation for female patients), custom-made pre-sternal implants that are designed to reduce the defect, external vacuum devices, corrective sternum osteotomy, various modifications of the Ravitch procedure and also the minimally invasive surgery introduced by Donald Nuss.

In 1998, Donald Nuss [4] published the results of a new technique, which he developed, currently referred to as minimally invasive repair of pectus excavatum (MIRPE). This operation elevates the depressed sternum by passing a suitably shaped bar beneath the sternum, resting on the outer aspects of the ribs on each side. The bar, or bars, are inserted under thoracoscopic guidance, through small incisions in the lateral chest wall, and secured in place by stabilisers that limit the potential for displacement.

Such a 'minimally invasive' approach has proven to be extremely popular amongst patients and thoracic surgeons,

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Table 1. Presentation of studies and reported results post-MIRPE.

Study and institution	Journal and year	Sample size	Indication for surgery[a]	Age at MIRPE	Surgeon cosmetic[b]	Patient cosmetic[b]	Self-esteem	Social life	Reported results following surgery				
									Lung capacity	Cardiovascular capacity	Exercise capacity	General health	SSQ[c] total score
Nuss et al.; Norfolk	J Pediatr Surg 1998	30	P	1.5–15	100%								
Jacobs et al.; Montreal	Eur J Cardiothorac Surg 2002	31	B	4.4–31.0	100%								
Hosie et al.; Mannheim	Eur J Pediatr Surg 2002	172	B	10.5–19.7	81.50%								
Croitoru et al.; Norfolk	J Pediatr Surg 2002	296	B	2.0 – 29.0	99.70%				↑	↑			
Borowitz et al.; Buffalo	J Pediatr Surg 2003	10	C	10.4–16.4					↔		↔		
Lawson et al.; Nolfok	J Pediatr Surg 2003	19	B	8.0–18		↑	↑	↑			↑	↑	
Sigalet et al.; Calgary	J Pediatr Surg 2003	11	P	10.4–16.6					↓	↔	↑		
Park et al.; Chunan	Ann Thorac Surg 2004	322	B	1.5–46	98.80%								
Krasopoulos et al.; London	Eur J Cardiothorac Surg 2005	20	C	14–37		80%	↑	↑			↔	↔	65
Kim et al.; Seoul	Ann Thorac Surg 2005	51	B	1.5–51		75%							
Bawazir et al.; Calgary	J Pediatr Surg 2005	48	B	11.8–15.2		↑			↔	↔	↑		
Lawson et al.;	J Pediatr Surg 2005	20	B	< 11					↔				
Nolfok		25		≥11					↑				
Schalamon et al.; Graz	J Thorac Cardiovasc Surg 2006	43	C	18–39	91%				↔		↔		
Coln et al.; Dallas	J Pediatr Surg 2006	107	P	5.0–18						↑			
Hebra et al.; St Petersburg	Am Surg 2006	30	C	18–32		100%							
Petersen et al.; Hannover	Eur J Pediatr Surg 2006	57	C	5.0–20		92%							
Kubiak et al.; Basel	Eur J Pediatr Surg 2007	15	B	10.7–18.1					↑				
Kelly et al.; Multicentre	Am Coll Surg 2007	284	B	3.0–21		85%							
Aronson et al.; Amsterdam	World J Surg 2007	53	C	6.1–32.1	98%			↑	↔		↑		
Metzelder et al.; Hannover	Ann Thorac Surg 2007	40	C	16–20		96%	↑	↑			↑	↑	67
Bohosiewicz et al.; Katowice	Eur J Pediatr Surg 2007	66	C	1.0–19	84%	100%							
Sigalet et al.; Calgary	Pediatr Surg Int 2007	26	B	11.1–14.3		100%			↑	↔	↑		
Nakagawa et al.; Okayama	J Pediatr Surg 2008	150	C	3.0–42	100%								
Lam et al.; Vancouver	J Pediatr Surg 2008	11	B	13.2–17.6		100%	↑					↑	
Kelly et al.; Nolfok	Pediatrics 2008	247	B	8.0–21		97%	↑	↑			↑		
Cheng et al.; Taipei	Eur J Cardiothorac Surg 2008	96	P	18–42		91.60%							
Hurme et al.; Turku	Scand J Surg 2008	25	B	5.0–23	92%							↑	
Pilegaard et al.; Sk. Aarhus	Inter CVT Surg 2008	73	C	7.0–43	100%								
Pilegaard et al.; Sk. Aarhus+Odense	Ann Thorac Surg 2008	180	C	18–43	99.50%								
Nuss & Kelly; Nolfok	Adv Pediatr 2008	947	B	<1–36+	↑	>95%							
The et al.; Rochester	Ann Thorac Surg 2008	19	B	17–42		100%					↑		
Al-Assiri et al.; Calgary	J Pediatr Surg 2009	30	B	10.9–19.0	100%	↑			↔	↔	↑		
UK registry; 13 centres	NICE Int J Tech Ass Health Care 2010	260	B	5.0–85		↑							

↑: improved; ↓: deteriorated; ↔: no change.

[a] C: cosmesis; P: physiologic; B: both cosmesis and physiologic.

[b] Reported % of patients with cosmetic improvement.

[c] SSQ: Single Step Questionnaire.

Table 2. Presentation of studies and reported results post-removal of pectus bar(s) and completion of MIRPE.

Study and institution	Journal and year	Sample size	Age at MIRPE	Years between MIRPE and bar removal	Number of patients with bars removed	Post-bar removal recurrence (%)
Nuss et al.; Norfolk	J Pediatr Surg 1998	30	1.5–15	2	30	10%
Jacobs et al.; Montreal	Eur J Cardiothorac Surg 2002	31	4.4–31.0	2.0, 3.0, 4.0	30	2%
Croitoru et al.; Norfolk	J Pediatr Surg 2002	296	2.0–29.0	<1 to >5	71	9%
Jo et al.; Seoul	J Korean Med Sci 2003	15	2.0–25	2	15	7.70%
Park et al.; Chunan	Ann Thorac Surg 2004	322	1.5–46	2	44	0%
Kim et al.; Seoul	Ann Thorac Surg 2005	51	1.5–51	1.5–4.5	36	0%
Bawazir et al.; Calgary	J Pediatr Surg 2005	48	11.8–15.2	2	11	0%
Lawson et al.; Norlfok	J Pediatr Surg 2005	55	Median: 11.4	3	55	0%
Schalmon et al.; Graz	J Thorac Cardiovasc Surg 2006	43	18–39	3	15	20%
Dzielicki et al.; Zabrze	Eur J Cardiothorac Surg 2006	461	<3 to >25	2.0–2.5	260	3.30%
Kubiak et al.; Basel	Eur J Pediatr Surg 2007	15	10.7–18.1	3	15	0%
Aronson et al.; Amsterdam	World J Surg 2007	53	6.1–32.1	2	53	0%
Metzelder et al.; Hannover	Ann Thorac Surg 2007	40	16–20	2	40	2.50%
Bohosiewicz et al.; Katowice	Eur J Pediatr Surg 2007	66	1.0–19	2	24	4.50%
Sigalet et al.; Calgary	Pediatr Surg Int 2007	26	11.1–14.3	2	26	0%
Nakagawa et al.; Okayama	J Pediatr Surg 2008	150	3.0–42	1.9–3.0	150	0%
Cheng et al.; Taipei	Eur J Cardiothorac Surg 2008	96	18–42		7	0%
Hurme et al.; Turku	Scand J Surg 2008	25	5.0–23	1.0–3.0	8	13%
Pilegaard et al.; Sk. Aarhus	Inter CVT Surg 2008	73	7.0–43		73	1%
Pilegaard et al.; Sk. Aarahus + Odense	Ann Thorac Surg 2008	180	18–43		46	0%
Nuss and Kelly; Norfolk	Adv Pediatr 2008	947	<1–36+	2.0–4.5	521	3%
The et al.; Rochester	Ann Thorac Surg 2008	19	17–42	2	6	33%
Al-Assiri et al.; Calgary	J Pediatr Surg 2009	30	10.9–19.0	2	30	0%
UK registry; 13 centres	NICE Int J Tech Ass Health Care 2010	260	5.0–85	2.7	81	0%

despite limited reports on outcomes. Given that the primary motive for most patients seeking surgical correction is concern for their physical appearance and its social acceptability, it is not surprising that smaller scars located away from the most visible areas of the chest wall are most preferred [5].

A large number of institutions have reported their results with MIRPE, but few have reported outcomes that are relevant to the concerns, which led the patient to undergo such surgery, and even fewer give long-term results following bar removal. By summarising the data in the literature, we hope to be able to provide clinicians and patients with a valuable resource of information and help patient choice.

In this article, we address the effectiveness of MIRPE, the sustainability of any improvement and identify areas that require further research.

2. Methodology

We have searched Medline and EMBASE applying the Ovid interface, using the following keywords: 'results', 'outcome', 'MIRPE' and 'Nuss procedure.' The search revealed 179 hits, which were independently assessed and led to a formal review of 80 publications. We have excluded all studies with less than 10 patients, as we thought that these might represent the surgical learning curve, and as such might not be representative.

Thirty-five studies were found. They reported results from the perspective of the patients and/or surgeons and were included in this review. There were a number of publications that reported results from the same institution; 18 reports from five institutions, two to eight from each institution. We retained these publications as they frequently provide additional information regarding postoperative improvement but only the largest series from each institution was used when calculating the total number of patients who underwent MIRPE. We also included data from the 'MIRPE registry of the United Kingdom' [41], which had initially reported results on the website of the National Institute of Clinical Excellence (NICE) in 2009 (http://www.nice.org.uk/nicemedia/live/11087/45258/45258.pdf). Thirty-three studies reported results after MIRPE with the bars *in situ* (Table 1), and 24 studies reported on patients, who had had their bar(s) removed (Table 2).

Table 3. Levels of evidence (LOE), as per Harbour R and Miller J.

Levels of evidence (LOE: 1–4)	
1++	High quality meta-analyses, systematic reviews of RCTs, or RCTs with a very low risk of bias
1+	Well conducted meta-analyses, systematic reviews of RCTs, or RCTs with a low risk of bias
1–	Meta-analyses, systematic reviews or RCTs, or RCTs with a high risk of bias
2++	High quality systematic reviews of case control or cohort studies or high quality case control or cohort studies with a very low risk of confounding, bias or chance and high probability that the relationship is causal
2+	Well conducted case control or cohort studies with a low risk of confounding, bias, or chance and a moderate probability that the relationship is causal
2–	Case control or cohort studies with a high risk of confounding, bias, or chance and a significant risk that the relationship is not causal
3	Non-analytical studies, e.g. case reports, case series
4	Expert opinion

Table 4. Grades of evidence (LOE), as per Harbour R and Miller J.

Grades of recommendations (Grade: A—C)	
A	At least one meta-analysis, systematic review, or RCT rated as 1++ and directly applicable to the target population or A systematic review of RCTs or a body of evidence consisting principally of studies rated as 1+ directly applicable to the target population and demonstrating overall consistency of results.
B	A body of evidence including studies rated as 2++ directly applicable to the target population and demonstrating overall consistency of results Or Extrapolated evidence from studies rated as 1++ or 1+.
C	A body of evidence including studies rated as 2+ directly applicable to the target population and demonstrating overall consistency of results Or Extrapolated evidence from studies rated as 2++.
D	Evidence level 3 or 4 or Extrapolated evidence from studies rated as 2++.

Post-MIRPE results were assessed and ranked according to the levels of evidence (LOE: 1—5, Table 3) [40]. Recommendations made in this article are explicitly linked to the supporting evidence of the included literature and graded according to the strength of that evidence (Grade: A—D, Table 4) [40]. This grading system provides an estimate of the size of the treatment effect and an estimate of the certainty of the treatment effect. It is important to note, however, that level B or C of evidence does not imply that the recommendation is weak.

3. Results

All of the reports included in this review were published between 1998 and 2010. Thirty-two [4,6—36,38] publications with 2997 patients (median: 53; range: 10—947) reported results following MIRPE with the metallic bars *in situ* (Table 1). Twenty-three publications [4,6,11,13—17,21,23—27,30—38] with 1393 (median: 36; range: 6—521) patients reported results after MIRPE completion with the bar(s) having been removed (Table 2). One study reported the results of MIRPE in 50 patients in whom a previous, conventional repair of PE had failed (redo surgery) [27]. Of the 50 patients who have had a secondary procedure, 27 had undergone a prior Ravitch procedure, 23 had undergone a prior MIRPE and two of the prior Ravitch patients had also undergone a prior Leonard procedure.

In addition, retrospective data was included from the UK registry [41] on 260 cases collected from 13 institutions over a 9-year period (2000—2007) and also published on the NICE website (http://www.nice.org.uk/nicemedia/live/11087/45258/45258.pdf). The mean age of these patients was 16 years (5—85 years) and the majority were males (88%).

3.1. Indications for surgery

Indication for surgery (Table 1) was: mixed (cosmetic and/or physiological) for 19 publications with 2185 patients; cosmetic only for 11 publications with 609 patients; and physiological for four publications with 244 patients. We can safely say that from 32 studies, all but four offered MIRPE on cosmetic grounds.

A number of authors [2—4,7,10,11,13—15,17,19,21,22,25,26,28—31,34—40] have measured Haller's index or pectus severity index (PSI) as part of their preoperative assessment. Although a PSI of >3.2 was used as an inclusion criterion for MIRPE in some reports, in some of these, the authors admitted that they had performed MIRPE upon patients with a PSI of <3.2 [4,11,13,14,17,25,28,30,36,38,27,39]. Twelve publications [5,6,8,9,12,16,18,20,23,24,32,33] did not report any PSI measurements.

3.2. Age at time of MIRPE

It is evident from the literature that MIRPE has been implemented in three different groups of patients, one paediatric (age at operation: less than 11 years), one adolescent (age at operation: between 11 and 18 years) and one with adults patients (age over 18 years at operation).

Eighteen studies [6,7,11,13,15,20—23,25,27,29,31,32,34,36,39,41] (Table 1) report from a mixed population from all three age groups. Five publications [4,8—10,18] report from a mixed paediatric and adolescent population. Three publications [14,26,28] report on a population of only adolescent patients. Three [12,24,35] publications report on a mixed population of adolescent and adult patients and the last four [16,19,30,33] on a population of only adults. It would be rather inappropriate to combine outcomes from all of these groups or extrapolate results from one group to another.

3.3. Improvement in cosmetic appearance, assessed by the surgeon (LOE: 4, Grade D)

Fourteen publications on 2019 patients reported cosmetic outcome assessed subjectively by the operating surgeons (Table 1). The results were reported as 'improved', 'excellent', 'very good', 'good' and 'poor/failed.' In this report, we have merged the 'excellent' or 'very good' and 'good' results and we report them as 'improved.' The surgeons reported 76—100% of the operated patients with improved cosmetic outcomes following MIRPE.

3.4. Improvement in cosmetic appearance, assessed by the patients (LOE: 2++ and 2+, Grade: B)

Seventeen studies reported cosmetic outcome assessed by the patients, or in the case of the paediatric groups, by parents (Table 1). Different methods were used to determine

Table 5. Presentation of studies and reported results for lung volumes following MIRPE and MIRPE completion with the metallic bars removed (% predicted).

Study and institution	Sample size	Age at MIRPE	Test time post-MIRPE (months)	FVC%				FEV_1%				FEF_{25-75}%				TLC%			
				Preoperative		Postoperative		Preoperative		Postoperative		Preoperative		Postoperative		Preoperative		Postoperative	
				Median (IQR/R*)	Mean (SD**)	Median (IQR/R*)	Mean (SD**)	Median (IQR/R*)	Mean (SD**)	Median (IQR/R*)	Mean (SD**)	Median (IQR/R*)	Mean (SD**)	Median (IQR/R*)	Mean (SD**)	Median (IQR/R*)	Mean (SD**)	Median (IQR/R*)	Mean (SD**)
Croitoru et al.; Norfolk	296	2.0–29.0	Early																
Borowitz et al.; Buffalo	10	10.4–16.4	6–12	91 (62–108)		86 (49–110)		91 (65–112)		88 (57–116)		93 (66–120)		100 (58–131)		91 (73–114)		91 (62–112)	
Sigalet et al.; Calgary	11	10.4–16.6	3		92 (22)		72 (15)		79 (22)		68 (19)						108 (24)		96 (16)
Bawazir et al.; Calgary	10	11.8–15.2	Completion ***		88.66 (2.9)		86.5 (3.9)		79.19 (2.42)		80.4 (4.09)						98.21 (2.86)		98.1 (5.04)
Lawson et al.;	20	<11	Completion	86 (75–94)		89 (79–99)		86 (76 –94)		88 (79–97)		86 (69–102)		86 (70–93)					
Norfolk	25	≥11	***	84 (76–92)		90 (78–101)		81 (74–90)		90 (82–100)		77 (63–89)		91 (81–103)					
Schalamon et al.; Graz	43	18–39	6																
Kubiak et al.; Basel	15	10.7–18.1	Completion ***	63.7 (33–125)		81.3 (64–102)		62.9 (36–116)		84.7 (54–112)		58.2 (38–121)		71 (28–163)		83.2 (47–139)		88.3 (52–98)	
Aronson et al.; Amsterdam	53	6.1–32.1	Completion ***		89.4 (11.53)		89.26 (12.41)		94.91 (13)		95.19 (14)		88.45 (22.37)		91.04 (22.79)		90.13 (8.77)		89.19 (10.62)
Sigalet et al.; Calgary	26	11.1–14.3	Completion ***		89.5 (18.6)		92.4 (20.6)		78.40 (16)		84.2 (18.4)						95.3 (16)		99.3 (13.7)
Al-Assiri et al.	15	11.6–15.4	Completion		87.73 (14.34)		89.47 (16.18)		78.13 (10.41)		80.07 (9.2)						95.4 (15.78)		97.13 (13.25)
Calgary	15	10.9–19.1	***		89.53 (0.94)		92 (22.64)		79.3 (18.91)		85.6 (20.78)						94.33 (18.29)		97.2 (17.85)

* Inter-quartile range or range.
** Standard deviation.

this outcome, from grossly reported or scaled changes to detailed prospective analysis of specific questionnaires. Seven studies [9,13,20,22,25,29,34] assessed cosmetic results from a mixed population with paediatric patients included into the analysis, where a direct or indirect parental input can be a major confounding factor, and they have reported an 85–100% satisfactory cosmetic outcome. In one of these studies, Kelly et al. [22] reported 85% satisfied patients and 95% satisfied parents. Three studies [14,26,28] reported results from adolescent populations with 100% satisfactory cosmetic results. Five studies [12,24,30,35,36] reported results from a mixed adolescent and adult population with 80–100% satisfactory cosmetic outcome and one study [19] reported 100% satisfaction from an adult population. The UK registry [42] reported changes of the patients' assessed 'cosmetic appearance' (scale: 1–10) from a preoperative mean of 3.1 to a mean of 8.4, following MIRPE (for 119 out of 260 operated patients; median follow-up: 170 days).

3.5. Improvement in self-esteem (LOE: 2++, Grade: B)

Five studies reported changes in the self-esteem of the operated patient with a prospective usage of specific questionnaires (Table 1). Two studies used the Single Step Questionnaire (SSQ) [12,24] upon a population of adolescents and adults; two used the Pectus Excavatum Evaluation Questionnaire (PEEQ) [9,29] upon a mixed population of paediatric, adolescents and adults; and one study used a combination of the Child Health Questionnaire (CHQ-CF87) and the PEEQ [28] upon a population of adolescent patients. All of them reported a large percentage of patients (96–100%) with documented improvement to their self-esteem following MIRPE.

3.6. Improvement in social behaviour (LOE: 2+ and 2−, Grade: C)

Five publications reported on positive changes in patient social life and social behaviour post procedure (Table 1). Two studies used the SSQ [12,24] on a population of adolescents and adults and two used the PEEQ [9,29] on a mixed population of paediatric patients, adolescents and adults. One study [23] simply reported on positive changes in an adolescent population. We noted a large variability among patients, with 0–40% of those asked reporting no changes when assessed by the SSQ and with deployment of a specific question addressing this issue. When PEEQ was used on patients to assess psychosocial changes and of the nine psychosocial questions asked, all but one item showed significant improvement after surgery. Similarly, their parents confirmed significant improvements on all psychosocial items and also conveyed a significant reduction in their own concerns regarding the effects of PE on their child's life.

3.7. Assessment of lung capacity (LOE: 2+ and 2−, Grade: C)

Four studies [8,10,16,38] reported on changes in lung function parameters following MIRPE and six studies following MIRPE completion [14,15,21,23,26,36] (Table 5). Although there has been variation in the types of lung volumes tested amongst the studies, the most consistent findings related to the following:

- Forced vital capacity (FVC);
- Forced expiratory volume at 1 min (FEV_1);
- Forced expiratory flow from 25% to 75% (FEF_{25-75}) of the vital capacity (VC); and
- Total lung capacity (TLC).

Schalmon et al. [16] reported no statistical significant changes at 6 months post-MIRPE from 20 patients while Borowitz et al. [8] reported no significant changes for FVC, FEV_1, FEV_1/FVC, FEF_{25-75} and TLC, from 10 patients at 6–12 months post-MIRPE.

Two studies included different populations from the same institution in Norfolk, USA [15,38]. Lawson et al. [15] reported a statistically significant improvement in FVC (6%, inter-quartile range (IQR): 2–9%), FEV_1 (9%, IQR: 8–10%), FEF_{25-75} (15%, IQR: 14–18%) for 25 adolescents ($\geq$11 years) but no improvement on the same tests for 20 children ($<$11 years) post-MIRPE completion. The average time for both groups, between bar removal and the postoperative pulmonary function tests (PFTs) used in this analysis, was 1.2 years (range, 0.1–3.8 years). Croitoru et al. [38] reported improvement in pulmonary function (no clear determination of the tests, mixed population: 2–29 years) in 117 of 163 patients (72%) tested.

Four articles were published from Calgary, Canada [10,14,26,36], which appear to be contradictory. The first article by Sigalet et al. [10] had shown a deterioration for each test in FVC ($-18\% \pm 18$), FEV_1 ($-8\% \pm 13$) TLC ($-10\% \pm 13$) and VC ($-16\% \pm 16$) for 11 adolescent patients at 3 months post-MIRPE. In a follow-up article, Sigalet et al. [26] advocated a statistically significant improvement in FVC, FEV_1 and diffusion capacity of 26 patients post-MIRPE completion, while an earlier publication by Bawazir et al. [14] and a later article by Al-Assiri et al. [36] from the same group reported no statistically significant change in FVC, FEV_1 and TLC after MIRPE completion in a total number of 40 patients.

Aronson et al. [23] have mainly shown a statistically significant improvement for FEF_{25-75} (mean: $4.71\% \pm 4.65$) at 6 months post-MIRPE, which was lost after MIRPE completion. The rest of the volumes tested have shown no significant changes either at 6 months post-MIRPE or post-MIRPE completion.

Kubiak et al. [21] have shown a statistically significant improvement (with very large ranges) for FVC (38.4%, IQR: 1.7–59.4), FEV_1 (40.4%, IQR: 2.6–64.1) and residual volume/total lung capacity (RV/TLC) (−32.7%, IQR: 45.7–24.2) from 15 patients post-MIRPE completion.

3.8. Changes in cardiovascular parameters (LOE: 2+ and 2−, Grade: C)

In three publications [10,14,38], the authors considered that cardiovascular parameters had shown improvement following MIRPE, whilst in three others [18,26,36] there was thought to be no change (Table 1). Differences in the populations under study, significant differences in the

methodology and the fact that the same institution reported contradicting results [10,14,26,36] on different occasions make it difficult to understand these discrepancies.

Croitoru et al. [38] reported on a mixed population of children, adolescents and young adults (296 operated patients, with six failures). They reported improvements in cardiac compression in 98.1% of patients when assessed by echocardiography or computed tomography (CT). From the 141 patients with preoperative conduction problems, 117 (83%) patients had documented resolution. Out of the 48 patients with mitral valve prolapse (MVP), 20 had post-operative evaluation and 9 of 20 (45%) had documented resolution of MVP by ECHO. The authors did not specify at what point in time these assessments were made post-MIRPE.

Sigalet et al. [10] have shown a statistically significant improvement in stroke volume (SV: from 61.6 ± 24 to 77 ± 23 ml) from patients aged 10–16 years of age, who were evaluated by ECHO preoperatively and at 3 months following MIRPE. Interestingly, the same study reported no statistically significant change in cardiac output (CO) or cardiac index (CI) over this period. Bawazir et al. [14] investigated the impact of MIRPE in 48 adolescent patients. They have shown a statistically significant improvement in CO (4.79–5.64) and CI (3.34–3.79), but not of the SV at 3 months post-MIRPE. However, 22 patients assessed at 21 months post-MIRPE and 11 patients assessed after they had their bar(s) removed have shown no statistically significant changes in any of the above parameters. On a later study by Sigalet et al. [26] 26 adolescents who had SV, CO and CI assessed preoperatively as well as post-bar removal were found to have a statistically significant positive change in SV (from 69 ± 21.2 to 83.9 ± 24.5) and CO (from 4.66 ± 1.39 to 5.38 ± 1.48) but not for CI. They concluded that the changes in SV and CO 'may have been associated with the general growth of the patients.' Al-Assiri et al. [36] investigated 30 adolescents preoperatively and 3 months after MIRPE completion with the bar removed. They also failed to show any statistically significant changes to any of the cardiovascular parameters (SV, CO and CI) post-MIRPE completion.

Coln et al. [18] used exercise ECHO and electrocardiogram (ECG) to assess changes in cardiovascular parameters and demonstrated that cardiac compression was present in 117 (95%) patients preoperatively, mitral valve abnormality was detected in 54 (44%) patients (prolapse: 25 and regurgitation: 29) while six patients had significant arrhythmias. MIRPE led to a 92% improvement in cardiac compression (100 patients, 93% of those studied) and no patients had cardiac arrhythmias postoperatively. Seven patients had persistent MVP after surgery and one had mild mitral regurgitation. It is important to note here that all subjects included in this study were cadiovascularly asymptomatic prior to and also following the PE correction.

3.9. Improvement in exercise capacity (LOE: 2+ and 2–, Grade: C)

Twelve publications assessed post-repair exercise capacity with two studies reporting results from the same institution in Norfolk, USA [9,29] and four from the same institution in Calgary, Canada [10,14,26,36].

Four of them used prospectively applied questionnaires [9,12,24,29]. In two studies which used the SSQ (with a direct question assessing this domain), the authors found contradictory outcomes; one study [12] reported no changes in exercise capacity following MIRPE and the other [24] reported an improvement following MIRPE, which was sustainable and even increased following MIRPE completion. In the other two studies [9,29], where the PEEP questionnaire was deployed, the authors reported a statistically significant reduction in the mean obtained from the section regarding physical difficulties (three questions for patients and five questions for parents), from 2.14 ± 0.75 to 1.32 ± 0.39.

Four studies concentrated upon changes in VO_{2max} ($1\ kg^{-1}\ min^{-1}$) and anaerobic threshold. One study [9] reported an early deterioration at 3 months post-MIRPE (VO_{2max}% expected: -0.42 ± 25, 11 adolescents). A second study [8] reported no changes in VO_{2max} after 6–12 months (10 adolescents). A third study [14] reported a statistically significant deterioration in the VO_{2max}% expected at 3 months (32 adolescents; from 21.41 ± 1.78 to 18.4 ± 1.18) with improvement at 21 months (20 adolescents; from 21.41 ± 1.78 to 24.51 ± 2.39) and post-MIRPE completion (10 adolescents; from 21.41 ± 1.78 to 22.62 ± 2.1). The fourth study [26] reported no changes in the VO_{2max} post-bar removal (26 adolescents). One publication [36] reported a statistically significant change in the patients' ability to sprint with no changes in VO_{2max} (30 adolescents).

Two publications had poorly defined means of assessing exercise capacity, reporting it as either 'improvement' [38] or 'no change' [16] with no supporting data.

3.10. Improvement in 'general health' (LOE: 2+ and 2–, Grade: C)

Five studies reported an overall improvement (35–100%) in the 'general health' of the operated individuals. Two studies used the SSQ [12,24] on a population of adolescents and adults; one used the PEEQ [9] on a mixed population of paediatrics, adolescents and adults; one study used a combination of the CHQ-CF87 and the PEEQ [28] on a population of adolescent patients; the fifth study reported subjective data, for example, 'feeling better' post-MIRPE [31]. The variability in the improvement of 'general health' (35–100%) and the fact that most studies had included patients who underwent surgery for cosmetic reasons, highlights the fact that despite their young age, a large number of patients had very good 'general health,' which would be difficult to further improve.

3.11. Results from SSQ total score (LOE: 2++, Grade: B)

Two studies [12,24] reported on the total score obtained from the prospective implementation of the SSQ in a population of 60 adolescents and adults who had undergone MIRPE and 40 adolescents who had undergone MIRPE completion. The total score obtained from the SSQ enabled measurement of changes that occurred to both physiological and psychological parameters after MIRPE. Both publications showed a net gain on the 'total score', well above the 'lower satisfaction zone', for the vast majority of the patients following MIRPE and MIRPE completion.

3.12. Results following bar removal (LOE: 2++, Grade: B)

Twenty-three studies (Table 2) reported outcomes following the removal of metallic bar(s). Although a number of those publications had reported results from the same institution at different times or for different subgroups, after combining the largest representative studies from each institution, we ended up with a population of 2682 patients, out of which 1393 had their bars removed, representing 52% of the initial population. Eighteen studies reported removal of the metallic bar(s) after 1.9—4.5 years following the initial correction.

The mean reported recurrence post-removal of the metallic bar(s) is 5% (ranges: 0—33%). Five [4,16,31,35,37] small studies reported from small groups of 30 patients or less. These studies reported the highest, 8% overall rate of recurrence (range: 10—33%). However, if we only consider the 11 studies that had over 30 patients with their bars removed, and exclude three studies that reported results from similar institutions, a total of 1258 patients remain (90% of the total number). The reported mean incidence of recurrence in these larger and more reliable studies is as low as 1% (range: 0—3.3%).

The most frequently reported causes of recurrence were: weak bar (early reports), early removal of the bar (before 2 years) and MIRPE in patients with Marfan's syndrome (>20% chance of recurrence following MIRPE completion) [38]. Marfan patients who undergo MIRPE have a higher chance of developing pectus carinatum deformity due to overcorrection [38]. Two articles reported additional surgery [16,17] for 'removal of costal cartilages' at 20% and 3.3%, respectively. Removal of metallic bar(s) can be associated with a higher chance of reoccurrence in paediatric cases and within 6 months following MIRPE completion [25].

3.13. Results following redo-MIRPE for PE (LOE: 3, Grade: D)

Croitoru et al., Norfolk, USA [39], reported on 50 patients who underwent redo-MIRPE. Twenty-seven of these patients had undergone a prior Ravitch procedure and 23 had undergone a prior MIRPE. The median age at initial repair was 9.0 years (range: 1—19 years). Recurrence had occurred immediately or after 7 years following Ravitch and immediately or after 14 months following MIRPE. The median age at the time of redo surgery was 16.0 years (range: 3—25 years). The indication for redo surgery was reoccurrence of PE deformity in all cases. They reported a 95% improvement in subjective 'surgical team-based assessment' of cosmesis. Using an undetermined questionnaire, the authors also reported an 82% improvement in patient-based assessment of cosmesis and an 85% increase in exercise tolerance in a subgroup of 23 patients. In the same 23 patients, postoperative PFTs showed slight improvement in half of the patients and no change or a slight decrease in the remaining patients. Eleven out of 19 (58%) patients who had preoperative PFTs and who had had a prior Ravitch showed improvement in their postoperative FEF_{25-75}%. Eight out of 14 (57%) patients who had preoperative PFTs and who had had a prior Nuss showed improvement in their postoperative FEF_{25-75}%. Seventeen patients had their bars removed. Results were excellent in 47%, good in 41%, fair in 6% and failed in 6%. These results are similar to those reported following primary MIRPE, which suggests that, in experienced hands, redo-MIRPE following a failed Ravitch or MIRPE can be performed with a very satisfactory outcome.

4. Discussion

In the 1980s, Dr Donald Nuss developed a minimally invasive procedure for correction of PE deformity, the results of which were published in 1998 [4]. Since then, MIRPE has proven increasingly popular with surgeons and patients.

We have seen from the literature and also from this review that cosmesis is the main referring complaint for MIRPE in 91% of patients. This is particularly so in Western society where cosmetic appearance is of great cultural importance and the pressure to conform is a powerful factor in social acceptance and integration. Patients with severe pectus deformities often express low levels of self-esteem and increased levels of social-status anxiety, which can be alleviated or even reversed following MIRPE for the majority (96—100%) of patients.

Haller's index or PSI of >3.2 is usually considered to indicate a severe PE deformity [42]. However, a number of the articles in this review [4,11,13,14,17,25,28,30,36,38,27,39] included patients for MIRPE with a PSI <3.2 or included no assessment of PSI [5,6,8,9,12,16,18,20,23,24,32,33]. This may reflect the fact that eccentric or barrel-chest deformations can lead to a 'false PSI calculation' of less than 3.2.

It is important to understand that the absolute degree of surgical correction is not necessarily linked to the level of satisfaction reported from the treated patients. Neither is the degree of surgical correction related to an improvement in perception of bodily appearance or social-status anxiety post-MIRPE. In this article, we can see that cosmetic outcomes following MIRPE were judged to be unsatisfactory in up to 19.5% of cases when assessed by the surgeon and up to 25% when assessed by patients. Bohosiewicz et al. [26], who reported on a mixed population of paediatric and adolescent patients, reported a 90% satisfactory cosmetic result as judged by surgeons but this figure increased to 100% when the patients were asked to score their satisfaction. These findings highlight the fact that surgeons and operated patients (and their parents) have different values and standards when assessing postoperative results. Further, the reported percentage of unsatisfactory results from the surgeon's or patient's perspective is neither comparable nor interchangeable.

Kelly et al. [29] found that the objective severity of PE is unrelated to the physical and physiological difficulties that the patient encounters and that significant psychosocial improvements can be independent of the surgical type offered to correct the PE. In addition to this, a 96% satisfactory cosmetic result, as reported by Krasopoulos et al. [12], was met by a 100% improvement in patient's self-esteem, suggesting that improvement in self-esteem is not necessarily dependent on a perfect repair of the deformity.

This review tried to comprehensively present the results following MIRPE, and we conclude that MIRPE is very effective in restoring personal confidence and self-esteem,

both of which can help patients towards a better social integration. We have noted a large percentage of patients (up to 40%) reporting no changes in their social life post-MIRPE. This could be because changes in social life require time and may not be reflected when follow-up is short. It is also difficult to determine these changes when the patient is a child or adolescent (with a social behaviour that is under constant change and development) or when the reported changes are based on a third-party assessment (parental).

It is important to be able to measure and report on self-perception, self-esteem and social behaviour following MIRPE. To do so in our institution, we devised, validated and implemented a specific questionnaire for MIRPE, the SSQ [12]. This questionnaire has since been used by other institutions [24]. Both reported a substantial improvement in all parameters linked to self-perception, self-esteem and social behaviour following MIRPE.

There is evidence from the literature that both lung volumes [2,15,22] and cardiovascular function [2] in patients with PE are lower than in non-affected individuals of the same age group. However, there is debate as to whether aerobic fitness is compromised as a function of PE or is secondary to a sedentary lifestyle. MIRPE is associated with further deterioration in lung capacity at 3 months, which returns to preoperative values within 6 months [10,14]. Whether there is improvement beyond this period due to higher levels than were recorded preoperatively is not clear from the literature.

Any improvement in cardio-respiratory function following MIRPE could be a reflection of improved anatomy following surgery but could be equally due to cardio-respiratory reconditioning that occurs due to increased exercise and involvement with social activities. Objective assessment of cardio-respiratory improvement following MIRPE is affected by the age range of the studied populations and the impact of growth on younger subjects. Further confounding factors may be related to postoperative pain or the restriction imposed by the presence of the metal bar(s). It is reasonable, however, to assume that there is no evidence that MIRPE adversely affects lung volumes or cardiovascular capacity in the medium- to long term. Furthermore, the combination of positive attitude towards exercise and increased social integration, along with a potential improvement in the lung and cardiovascular physic of these patients, should be regarded as a successful 'combined' outcome for MIRPE.

A present recommendation [33] states that metallic bar(s) should stay *in situ* for at least 2 years for children and 3 years for adolescents and adults. Despite this, several surgical teams reported leaving the metallic bar *in situ* for at least 3 or 4 years. Literature suggests, however, that there is no significant difference in the degree of correction post-MIRPE for different age groups [27], and that removal of the metallic bar(s) within the first 12 months following MIRPE is followed by a 60% failure rate [33]. Results from this review show that when the bars were removed at 2—4.5 years post-MIRPE, the PE reoccurrence or failure rate for the larger studies was 1% (0—3.3%). However, it was not possible to establish if adults had more recurrences when the bar(s) were removed before 3—4 years.

From a single study, we can see that redo-MIRPE in experienced hands has been shown to offer results that are at least as good as first-time correction [39], suggesting that previous failed surgery is not necessarily a contraindication to MIRPE.

We can also see that all reported studies are of a low LOE (2++ to 3). Any attempt to proceed to a randomised control trial would most probably face recruitment difficulties due to the strong preference of patients to proceed with MIRPE against any other form of repair. However, it is possible to standardise the way patients are assessed post-MIRPE as well as the way results are reported.

There are limitations to this article. We tried to analyse and merge the data to the best of our capabilities, but we were limited by various confounding factors and limitations in the way data were reported in the literature. On many occasions, the reported outcomes were presented in small numbers, were incomplete or not specific enough to allow us to merge them in a more scientific way that could help us perform a meta-analysis. Despite this, we felt that there is value in consolidating the currently fragmented literature and as such help to guide clinical decision making and future research in the field of repairs for PE and MIRPE. We did not address the issue of surgical technique or associated complications as these are very well reported in the literature [4,34,43,44] and are beyond the scope of this article. It is important, however, to note that MIRPE is a surgical procedure with associated risk from general anaesthetic, something that should be at the forefront of any discussions with the patient and relatives.

5. Conclusion

In this review, we made a first attempt to examine the published results on MIRPE and provide a clearer insight as to what the goals may be for this operation.

There is a clear need for standardisation in the way results are reported in the literature and a socioeconomic analysis with regard to gains, benefits and costs related to MIRPE.

MIRPE is a well-established procedure that may improve the cosmesis and self-esteem of patients with this deformity. Direct or indirect improvement in other physiological parameters may also help the 'well-being' of these patients and their social integration.

Author contributions

GK was responsible for study design, statistical analysis, data interpretation, and manuscript drafting. PG has revised the manuscript; he is the guarantor for this article. His involvement was critical to every phase of this work. He had access to the data, and controlled the decision to publish. All authors read and approved the final manuscript.

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