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Full Length Research Paper

Effectiveness and Limitations of Minimally Invasive Repair of Pectus Excavatum- a 10 years' Experience

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Abstract

Pectus excavatum (PE) is the most common congenital deformity of the chest wall. Because of its heterogeneity, the repair isn't generally simple to perform even at the time of minimally invasive repair. The objectives of this study were to investigate the association between PE and the prognostic factors using the minimally invasive repair. (MIR). A total of 140 consecutive patients who were admitted to our institution for PE were assessed and included into the present study. Type of deformity, severity, age, sex, symptoms, Haller index, pre-and postoperative respiratory function tests, pre-and post-operative echocardiography, body mass index, bone mass, patients' quality of life pre-and postoperative, length of clinic stay and postoperative complications in addition long-term follow up were analyzed. Between January 2004 and December 2014, there were 130 patients underwent MIR (115 males, their mean age 22 ± 15 years. Median follow up was 6.2 years. Excellent results were obtained in 92% (n=120), 5% had less satisfactory results (n=6) and 3% (n=4) had unsatisfactory repair. The causes of unsatisfactory repair were: [1] Rib fractures in elderly patients (age >40 years) (n=2), [2] Incomplete correction due to high bone mass >3.5 kg (n=2), and [3] Complex asymmetric deformity (n=2). Reoperation for repair was done in 3 patients (2%). One patient had intraoperative cardiac arrest with successful resuscitation, who required admission into the intensive care unit (ICU) for 24 hours. Mean hospital stay was 4 days. 3% of the patients presented with minor complications. Patients satisfaction was significantly high (p<0.0001). Time adjusted pectus bar removal showed no recurrence of PE. MIR procedure for repair of pectus excavatum is a safe and an effective method. Although MIR is becoming the gold standard for the repair of PE, still few patients with complex deformities would not be suitable for this method. Age, type of deformity and higher bone mass are the most independent factors affecting the results regardless the sex.

Keywords: *Nuss procedure; pectus excavatum; chest wall deformity.*

Introduction

Pectus excavatum (PE) is the commonest chest wall congenital deformity; it constitutes about 90% of childhood chest wall deformities with an estimated incidence of 1 in 400 births and prevalence of 2.6% in children age from 7 to 14 years. PE affects 5 times more males than females. It is described by posterior depression of the lower third of the sternum. In the vast majority of the patients with serious deformation, pectoral muscles are accounted for to be underdeveloped (1). Despite the fact that PE happens most commonly as solitary disfigurement, up to 20% of patients may have other skeletal abnormalities, most prominently scoliosis (2). About one-quarter of the patients experience cardiopulmonary side effects. A large portion of the patients experiences mental disorders because of their chest wall deformation. The exact etiology of PE is still obscure, however its rate increments in patients with family history of chest wall intrinsic irregularities (3,4).

There is evidence to support that PE can alter physiologic function and decrease cardiopulmonary capacity, both at rest and with exertion. A new prospective study demonstrates that PE has an adverse effect on both cardiac and lung functions especially on the exercise capacity (5). The surgical management intends to adjustment the thoracic wall disfigurement and the increase of cardiopulmonary capacities; which prompts the improvement of the quality of life through the evacuation of the weight on the heart and lung (6,7). In 1998, an innovative strategy for repair of PE by utilizing a retrosternal bar without costal cartilage resection was described by Donald Nuss (8). This procedure was created as a minimally invasive procedure to be the highest quality level technique for PE management (9,10). One important issue in PE repair is the optimal timing of correction, which is yet to be clarified, although early repair has been reported to be associated with fewer complications, still, the golden age for correction is during adolescence years (11-13).

Asymmetric deformities, as well as the massive bone structure of the thoracic wall especially the sternum, is associated with more complications and less repair satisfaction (14-15). The aim of the present study is to lighten the lessons learned in the most recent decade and to demonstrate the challenges in various cases even in experienced hands. In this study, three cut points were identified to be endpoints as regard, the outcome of surgery, quality of life and recurrence.

Materials and methods

Data collection

Data of all PE cases were collected from our hospital records database, family doctors and pediatric hospital records, also direct patient's contacts. Data for age, gender, weight, length, BMI, BM, symptoms, degree of the deformity, complications, results, time of the operative procedure, length of hospital stay, quality of life after repair, and the recurrence rate were analyzed.

Follow up

It was arranged with each patient in the form of outpatients' visits in the first 4 years postoperatively. After PB extraction, the follow-up was performed by personal contact in a large portion of the cases or by post. A quality of life questionnaire was developed and modified using the WHO-questionnaire for QOL and was sent to all patients with post or e-mail.

Statistical analysis:

Data were expressed as mean ± SD or as count and percentage. Statistical analysis was performed using the Student's t-test and a P-value <0.05 was considered significant for all analyses. The reference values for RFTs indices were based on age, gender, height, and weight. Non-parametric values were compared by the Wilcoxon's signed sum rank test.

We analyzed the results from the SSQ and compared the results with those obtained by the NQ-mA test, which we considered as an adequately validated reference questionnaire. We used Wilcoxon signed rank test to determine the statistical significance of differences, with a <0.05 level considered as significant. Spearman's correlation coefficient was used to assess the correlation between the answers received on the test-retest study, the correlation between the total scores obtained from the post-operative NQ-mA and the SSQ, and finally, the correlation between the net gain in total scores from the NQ-mA and the SSQ. Spearman's correlation coefficient value of 1 indicates complete agreement. The SPSS software (IBM-SPSS Inc, Chicago, IL, USA, version 22) was used. A statistical difference with P <0.05 was considered significant.

Patients

Patients' selection and study design:

We analyzed the data of all cases who were seeking repair for their PE at our institution between January 2004 and December 2014. A total number of 140 patients was assessed. 10 cases were excluded due to incomplete data. According to their age; patients were divided into two groups. Group 1 consisted of 72 patients and they were operated on younger than 25 years old. Group 2 consisted of 58 patients in whom PE corrected after the age of 25 years. All patients received the usual preoperative work up in form of posteroanterior (PA) and lateral (LT) chest X-ray, chest computed tomography (CT). Few patients received chest magnetic resonance tomography (MRT), echocardiography (Echo) and respiratory function tests (RFTs). Haller index (HI) was calculated on CT or MRT and used to define the degree of deformity. The bone mass (BM) was calculated in patients with BMI > 30.

Six postoperative follow up was done in the following intervals: 2 weeks after discharge from the hospital, 3, 6, 12 and 24 months, then the last follow up was to determine the timing for pectus bar (PB) removal.

Operative techniques

NP was performed for all patients. Numerous recommendations from different specialists were thinking about prevention of PB rotation. All patients were operated on in the supine position with abduction & external rotation of both arms under single lumen intubation. A bilateral mid-axillary skin incision was done (length: 2–3 cm). One or more retrosternal convex metallic bars (pectus bars) were exactly measured and bent according to body size and inserted under the thoracoscopic guide. On both ends of the bar, two side stabilizers were fixed with metallic wire. The measurement was done as depicted by Donald Nuss, the length of the PB was 2 inches less the line from one axillary line to the other. PB was settled with two stabilizers on both sides utilizing metal wires. Only in severe deformities, we used to fix the bar with metal wires around the rib (3 points fixation).

Results

Patients' characteristics

From the patient's database of our center, 140 consecutive patients were admitted and evaluated for PE repair. 10 patients were excluded (3 patients were refused from our surgeon himself, 7 patients did not show up). MIR was done for 130 patients; their mean age was mean age 21 years (range 7 to 53 years). There were 115 males (88%) and 15 females (12%). Group I consisted of 72 patients, mean age 17 (range 7 - 25 years) with mean BMI of 18,4 (range 11,2-28,6), and the mean BM of 2,5 kg (range 2,1-3,8 kg). The mean HI was 3.8 in group I (range 3.1-4.8). Group II consisted of 58 patients, their mean age 22.5 in group II, range (26-53 years) with a mean BMI of 26,4 (range 21,2-36,4), and the mean BM of 3,2 kg (range 2,6-3,9 kg). The mean HI was 3.6 (range 2.9-4.7). Median follow up in all patients was 6.2 years. Patients' characteristics are given in table 1.

Echocardiography & lung functions tests

They were performed preoperative and 6 months after the operation. The mean FEV1 was 78% (range 64%-93%). The mean VC was 85% (range 65%-105%). Post-operative RFTs (FEV1, VC) showed better values in comparison to preoperative values in 78% (n=102), no change in 17% (n=22) and less values in 5% (n=7). Restrictive values were noticed in 65% (n=85) preoperatively, which was less than 12% (n=16) in the post-operative follow-up (p<0.001).

Preoperatively, ECG was done in 88% (n=115) of the patients and transthoracic echo in fewer patients 26% (n=34) who had changes on ECG or suffering from severe deformity. The postoperative cardiac examination was done with the 6 months follow up timing. The most common abnormalities found on ECG and or echocardiography were: [i] dislocated apex of the heart much to the left (56%) (n=74), [ii] right bundle branch block (25%), [iii] left bundle branch block (16%), [iv] extrasystoles (8%), and [v] Angina-like findings 1%. All the cardiac findings disappeared on ECG and echocardiography apart of the right bundle branch block which persisted in 2 patients (1.5%).

Operative results

Excellent results were obtained in 92% (n=120), 5% had less satisfactory results (n=6) and 3 % (n=4) had unsatisfactory repair. The reason for unsatisfactory repair was unable to correct the PE due to intraoperative rib fracture during the elevation of the sternum using the sword. In these 3 patients, the PE deformity was very severe, their age ≥ 35 years, and had a calculated BM > 3.5 Kg. Reoperation due to PB displacement or unsatisfactory results was done in 3 patients. There were 5 patients (4%) who developed postoperative pleural effusion (3 on one side and 2 in both sides. 4 of them needed a chest tube, none was re-operated on due to pleural effusion. There were 18 patients with pneumothorax between 1-5 cm postoperatively or after chest tube removal (14%). 3 patients needed chest tube insertion, and in 15 patients pneumothorax was resorbed after 24 hours using 3-4L of O2 supply on the nose. Other rare complications were observed in 11% (n=14) and are listed in the table (2). Mean hospital stay was 4 days in group I (range 3-6 days) vs 6 days in group II (range 5-11 days); one admission to the ICU was needed due to intraoperative ventricular arrhythmia of unknown origin, which was successfully converted using intraoperative electrical shock. Dramatic reduction of PB slipping, rotation, flipping or hinge point were noticed after using side stabilizers ($p<0.001$). Removal of PB was done in 22 in group I vs. 24 in group II. No recurrence of PE was noticed in all patients after removal of PB in both groups.

Patients' satisfaction

There were 130 questionnaires were sent to the patients, patients were asked to fill in and return it back either by mail or e-mail or fax. There were 112 questionnaires returned, but 8 were not suitable to analyze due to missed or undefined data. 104 questionnaires were analyzed and subjected to comparison in both groups. There was very high ($p<0.0001$) patients' satisfaction in group I vs. high satisfaction in group II ($p=0.022$). Patients with pain more than 6 months were 2 in group I and 4 in group II. No patient answered with yes to the question if he/she would not do the operation or recommend it for somebody else ($p<0.000I$). 95% (n=68) in group I answered with yes for good/normal body feeling postoperatively compared with 12.5% (n=9) preoperatively ($p<0.001$). In group II 88% (n=51) answered with yes for good/normal body feeling postoperatively comparing with 5% (n=3) preoperatively ($p<0.001$). Other items of satisfaction are summarized in table 3.

Table 1: Patients characteristics

Patients characteristics	Group I	Group II
Number	72	58
Age (years)		
Mean±SD	17±8,4	22,5±9,5
Median	16.9	29,5
Range	7-25	26-53
Gender		
Male	63	52
Female	9	6
Body length, cm (mean)	163	176
Body weight, Kg (mean)	58	67
Body mass index, kg/m^2 (mean)	16.5	21.5
Degree of deformity		
Mild	4	3
Moderate	60	42
Severe	8	7
Morphology		
Symetric	55	36
Asymetric	17	16
Combined	6	4
Haller Index	3.8	3.6
Thoracic scoliosis		
No	61	51
Mild	7	5
Moderate	4	2
Severe	0	0
FEV1	78%	73%
Cardiac dysmorphia on CT (number)		
No	42	36

Mild	15	10
Moderate	8	7
Severe	7	5

Table 2: Complications

complications	Group I	Group II
Pneumothorax	12	6
Hemothorax	1	0
Pleural effusion	3	2
Wound infection	0	3
Rib fractures	0	2
Re operation	1	2
Pericardial injury	1	0
Intra operative reanimation	0	1
Allergic reaction	1	2
Bar flipping	1	5
Stabilizer dislocation	1	0
Temporary Horner's syndrome	1	0
Wound seroma	2	3
Shoulder pain	2	4
Pectus excavatum recurrence	0	1

Table 3: Post-operative satisfaction

Satisfaction	Group I (n=72)	Group II (n=58)
Surgeons' satisfaction intra operative:		
Highly satisfied	63	38
Satisfied	12	14
Less satisfied	1	4
Not satisfied	2	2
Surgeons' satisfaction post-operative:		
Highly satisfied	58	40
Satisfied	15	11
Less satisfied	2	5
Not satisfied	3	2
Patients' satisfaction post-operative:		
Highly satisfied	67	47
Satisfied	8	6
Less satisfied	2	3
Not satisfied	1	2
Patients' satisfaction 2 years post-operative:		
Highly satisfied	65	49
Satisfied	4	6
Less satisfied	1	1
Not satisfied	2	2
Patients' satisfaction after pectus bar removal:		
Highly satisfied	74	55
Satisfied	0	0
Less satisfied	1	1
Not satisfied	3	2
Parents/relatives' satisfaction:		
Highly satisfied	57	39
Satisfied	12	2
Less satisfied	5	2
Not satisfied	4	0
undecided	0	15

Discussion

Pectus excavatum (PE) and carinatum are the most widely recognized thoracic cage anomalies. They often coincide with the vulnerable life phase of puberty, a period characterized by great physical, social, and emotional changes. PE either can be present at birth or may develop and deteriorate during childhood and adolescence (16). Over the last 2 decades, the MIR was commonly used all over the world replacing most of the conventional methods known. Nowadays it is the gold standard for the treatment of PE. Excellent results were obtained in 92% (n=120), 5% had less satisfactory results (n=6) and 3% had unsatisfactory repair (n=4). Our results were similar to that seen by Kelly et al., (2013) who stated that satisfactory results were recorded in 93.8% of cases and only 6.2% had worsening of their conditions (17). In our study, no worsening of the deformity was reported. Nevertheless, MIR has its limitations especially in case of complex deformity e.g. the combination of PE and carinatum, PE with one side deformity of the ribs, PE combined with costal margin deformity or PE in an elderly patient with large sized thorax and heavy bone structures of the sternum. The correction of the PE depends not only on the degree of deformity but also on the age. Donald Nuss presented his series with 10 years of follow up in 1998 with a very young patient population (1.5–15 years) (8). There were many modifications of the original Nuss procedure, some of them are useful and some are of no benefit.

We agree with authors who reported that stabilizing of both ends of the bar using side stabilizers are most important (18,19). We disagree with authors who suggest 3-point stabilization for each patient (3, 13), as this is in many patients with symmetrical deformity even if it is severe really not needed. We disagree with Pilegaard and Licht (20) about the use of shorter bars, as this was shown through other authors that it leads to higher incidence of rotation and flipping of the bar (21). Asymmetrical PE or complex thoracic wall deformities need not only correction of PE using the Nuss procedure but much more modifications and combined surgery. Most of the publication before 2010 did not address these difficulties, very rare discussion and nearly no consensus in the literature about how to manage complex deformity, especially in an older patient with heavy sternal bone density undergoing MINP. In such patient with the rigid and heavy sternum, losing the lever effect scenario after rib fracture is common. This is a challenging situation for the surgeon. The median age of our study group was 21 years. This was older than that seen by other authors, where the median age was 14 years and 15 respectively (18, 22). Still little was reported about limitations of MIR in elderly patients with asymmetric deformity and how to deal with it. In our experience, the best age for MERPE was between 14 to 25 years old, and especially in patients with less bony density and BMI. Younger patients (<12 years) who have no cardiopulmonary symptoms should be advised to wait with the correction of their PE to avoid a second operation for a bigger bar size after two years due to size mismatch with their rapidly growing thoracic cage.

Our results showed a mean HI of 3,8 which is less than reported by other authors but still high on itself. In our study, we found a significant correlation with the higher HI and the use of more than one PB. Also, there was a significant correlation with the higher HI and the presence of cardiopulmonary symptoms, which was noticed by other authors as well (22,23). In this study, we noticed that the higher the HI, the higher the incidence of cardiac symptoms. Cardiac symptoms were mimicking angina pectoris in one female patient aged 41 years old in whom the HI was 4,8. She developed signs and symptoms of angina pectoris due to compression of the right coronary artery, which were asses as a coronary artery closure. She underwent open cardiac surgery and bypass of the right coronary artery. The postoperative course after her cardiac surgery was uneventful but her symptoms persisted. On the advice of her cardiologist, she sought our institution for correction of her PE. After PE correction with using MIR, all her cardiac symptoms have gone and showed no any signs or symptoms of angina pectoris anymore.

We noticed a significant correction of the RFTs in most of the patients underwent MIR. Preoperatively, 45% of our patients showed restricted lung functions with less FEV1 than expected, which were corrected in most of the patients (87%) after the correction of the PE. Many young patients with FEV1 less than 95% are subjected to suffer from early deterioration of their RFTs in earlier ages than other individuals who have the same life conditions without PE. Therefore, one has to advise them for correction of PE even if the FEV1 and the VC are still in better conditions. Although the effect of MIR on the cardiopulmonary functions is still controversial, we noticed that most of the cardiopulmonary symptoms were relieved. The preoperative cardiopulmonary data showed significant improvement in the postoperative work up 6 months after the operation and later on, these findings were reported earlier (17). To proof these retrospective findings, prospective studies are needed. Use of shorter bars was described by Pilegaard, did not find much acceptance between surgeons with more experiences. Also, the 3-point fixation around the ribs and the sternum to avoid hinge point displacement is still unpopular because of its accompanying complications as well as little evidence of its benefit (24). The mean follow up of our study was 5.2 years, which is longer than reported before (23). Only one patient has pain after 9 months postoperatively, which reported to be new. All other patients showed no symptoms after 6 months postoperatively. Usually, we leave the PB for 3 years duration in younger patients (≤ 25 years) and 4 years in older patients. Moreover, no recurrence after PB removal was noticed over the follow-up period. In conclusion, MIR is the gold standard of care for patients suffering from symmetrical PE. In case of severe asymmetrical deformity especially in older patients, with high BM every care must be taken to avoid rib fractures and bar dislocations.

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