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Pectus Excavatum Repair in Adults: Indications and How To Do It

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Abstract
Purpose of Review The minimally invasive Nuss procedure has been effectively used for surgical correction of pediatric and adolescent pectus patients. In the past, the use of this less invasive technique was believed to be limited to young patients. This approach with technique modifications has been successfully extended to the treatment of even advanced aged adults. The presentation, evaluation and minimally invasive surgical treatment of adult pectus excavatum patient is reviewed.
Recent Findings Adult pectus excavatum patients may present with new onset or worsening of symptoms. Repair of the deformity has been shown to improve cardiopulmonary deficits and quality of life. The increased rigidity of the chest wall in adult patients makes elevating the sternum and supporting the repair with substernal bars more difficult. Technique modification to the original Nuss procedure including the use of forced sternal elevation, multiple support bars and improved bar fixation methods have allowed for successful correction of even advanced age adult pectus excavatum patients.
Summary of Findings The modified "Nuss" is a safe and effective alternative to open repair of pectus excavatum in adult patients. Repair of the deformity can improve cardiopulmonary function and symptoms.

Keywords Pectus excavatum · Modified Nuss · MIRPE · Bar fixation · FiberWire · Sternal elevation

Introduction

Pectus excavatum and carinatum are among the most common congenital chest anomalies, with an overall incidence of pectus excavatum (PE) in 23 of 10,000 births [1–4]. Pectus excavatum is more likely to occur in men than in women; however, PE in females is often underdiagnosed because breast tissue may obscure the defect [1, 5]. The cause of PE is unknown but may be from unbalanced overgrowth in the costochondral regions. Studies have shown that patients with asymmetric PE had shorter ribs on the more severely depressed side of the defect [6, 7]. 43% of patients with PE have a family history of the defect [7]. Pectus excavatum is thought to be of multifactorial inheritance, but the exact genes are unknown [8–10]. Pectus excavatum is associated with scoliosis and connective tissue disorders including Marfan Familial Syndrome, Ehlers–Danlos, and Noonan syndromes [1, 7, 9, 10].

Depending on the severity, presentation of PE ranges from a minor cosmetic issue to incapacitating cardiopulmonary symptoms [11, 12•, 13]. The inward displacement of the sternum can cause restrictive defects as well as compression to the right heart [11, 12•]. Symptoms may appear or show progression as a patient ages. Increased calcium buildup in the cartilage attachments of the anterior chest wall can result in decreased chest wall flexibility and may be once cause of late onset of symptoms [14, 15•, 16]. In one report, development of symptoms did not occur until the 4th and 5th decade in nearly half of adult patients

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studied and symptoms improved significantly after surgical repair [15•].

The cardiopulmonary effects of PE have long been discussed [17–19], and there are only a few reports evaluating adult patients [20•, 21]. Data supports that there is a negative cardiopulmonary impact on patients with PE due to the inward projection of the chest wall [12•, 22] which causes cardiac displacement into the left side of the chest and varying degrees of cardiac compression (Fig. 1). With significant compression to the right heart chambers, reduction in atrial filling and venous return can cause diastolic dysfunction and decreased cardiac output [12•, 21–23]. Surgical repair of the PE has been shown to relieve compression which allows for an increase in right heart chamber size and flow velocities, as well as improved cardiac output [12•, 15•, 22, 24••, 25] (Fig. 2). In PE patients ≥30 years, there was reported a mean increase in right ventricular output of 65% after surgical repair as assessed on intraoperative transesophageal echocardiogram [24••]. Others have also reported a significant improvement in cardiopulmonary function with increase in both VO_2 and O_2 pulse after PR correction [11, 20•]. Normalization of decreased cardiopulmonary function has also been shown in teenagers three years following repair of PE [26•]. A recent evaluation of adults (≥21 years) one year after Nuss repair did not find a significant improvement in the VO_2 max and longer follow up is in process [27•]. This study did show a trend towards improvement in the VO2 max however it is not known whether the adult with PE has the ability to normalize after repair like the adolescent patient.

The optimal surgical treatment of adult patients with PE has been controversial with some recommending the Nuss procedure be limited to pediatrics and adolescents [28, 29]. We present a review of evaluation and treatment of adult patients with PE including our preferred method of the modified Nuss ("Nuss") or a minimally invasive repair of pectus excavatum (MIRPE).

Patient Presentation and Evaluation

Many PE patients do not exhibit symptoms until adolescence or adulthood [14, 30]. Adult patients may also have notable progression of symptoms with aging as the chest wall becomes less compliant and compensatory mechanisms decrease [14, 15•, 23, 31]. A variety of symptoms have been described (Table 1) with the most common being exertional dyspnea and tachycardia, exercise intolerance, and chest pain [15•, 24••, 32–35•, 36, 37]. Surgical correction of PE may improve outcomes in these patients [38]. Psychosocial issues and body image are factors that also cannot be underestimated [39].

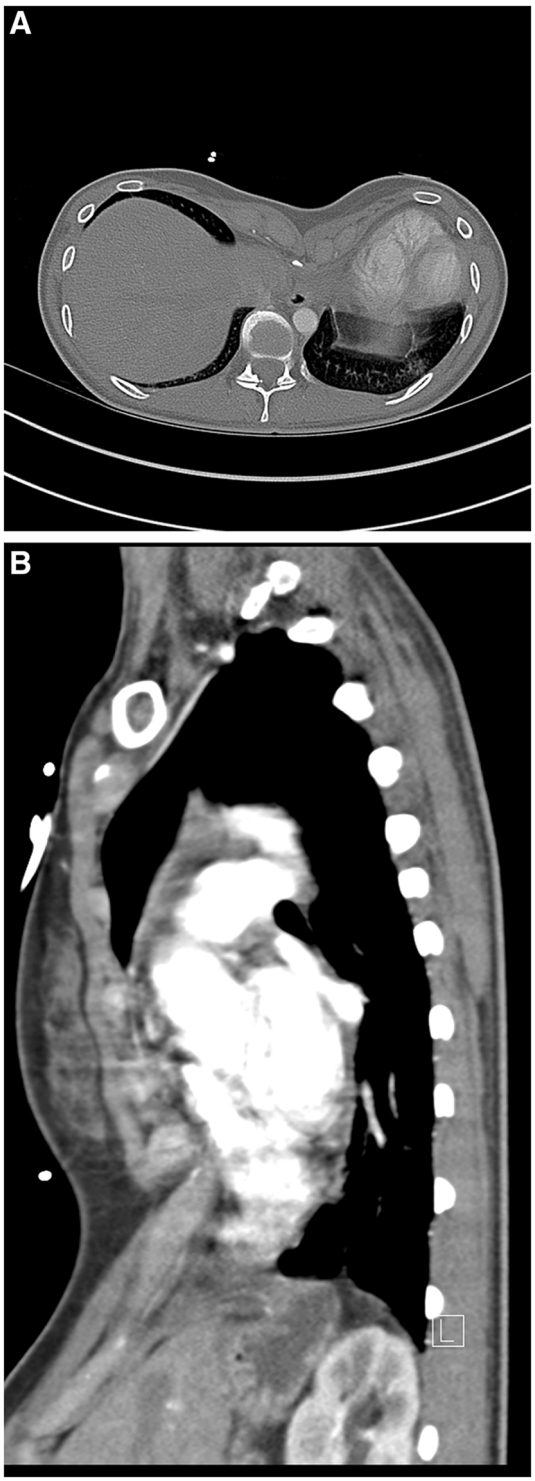

Fig. 1 **a** CT scan shows *left* heart shift. **b** Cardiac compression is visualized by CT

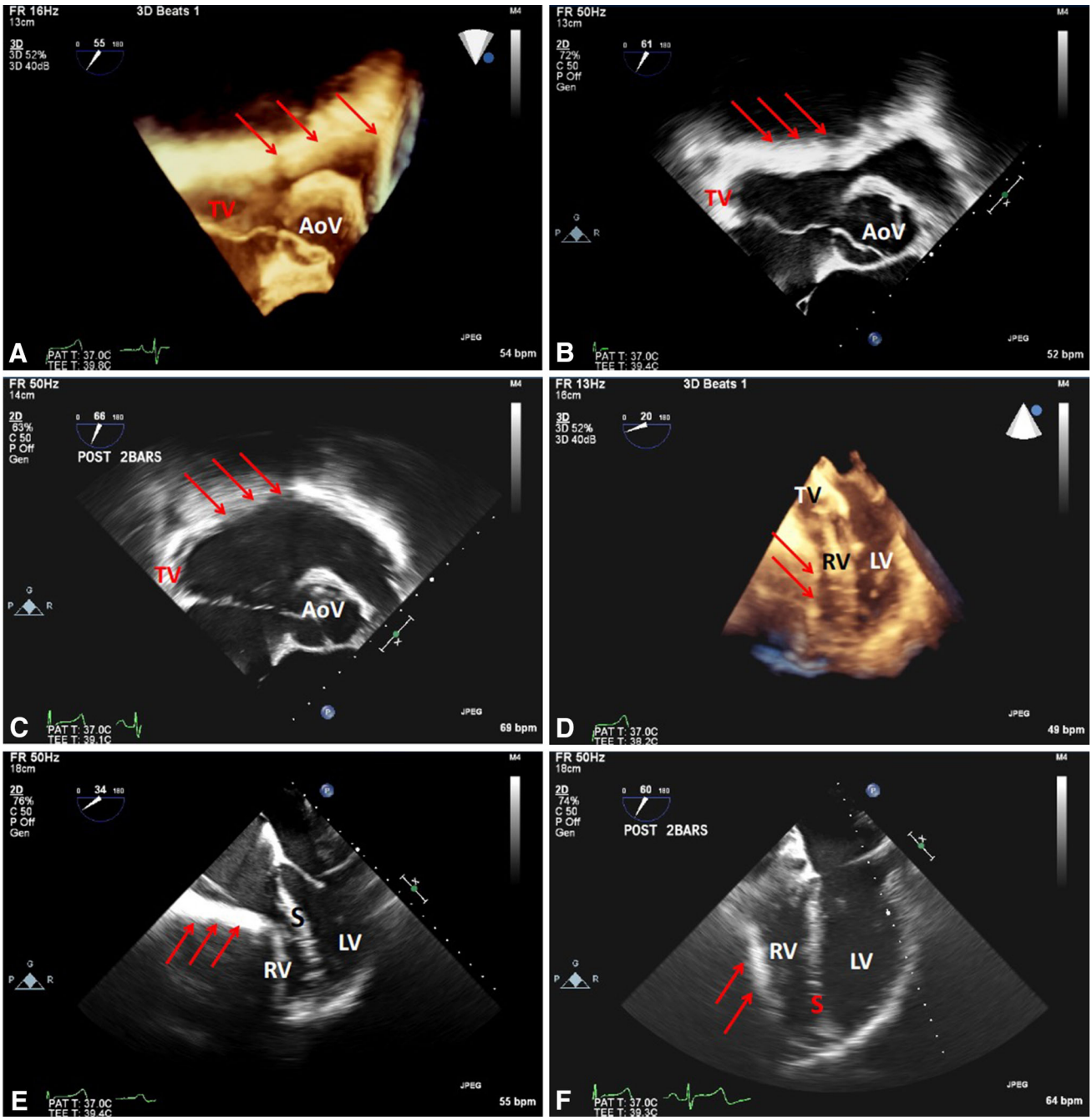

Fig. 2 **a** Transesophageal echocardiogram (TEE) 3D short axis image at the level of the aortic valve (AoV). *Arrows* indicate right ventricle outflow tract (RVOT) compression. **b** 2D TEE image based short axis at the level of the AoV. *Arrows* indicate RVOT compression. **c** 2D TEE short axis image at the level of the AoV post repair shows relief of compression. **d** 3D TEE four chamber view showing severe right ventricle (RV) compression (*arrows*). **e** 2D TEE four chamber view showing pectus compression (*arrows*) on the right ventricle. **f** Post-Nuss bar placement shows relief (*arrows*) of right ventricle compression. *TV* tricuspid valve, *LV* left ventricle, *S* ventricular septum

History and Physical Examination

When an adult patient presents with PE, an evaluation should be completed to understand the severity of the condition and to rule out other cardiopulmonary disorders. A thorough workup is indicated in patients with symptoms and can include history and physical examination; thoracic imaging, blood chemistry analysis, electrocardiogram, echocardiogram, cardiopulmonary exercise testing through measurement of peak oxygen uptake (VO_2), and pulmonary function tests.

Table 1 Common presenting symptoms of significant pectus excavatum

Exertional dyspnea
Prolonged exertional tachycardia
Palpitations
Fatigue
Poor exercise tolerance
Chest discomfort or pressure
Postural symptoms
Syncope
Dysphagia
Social anxiety related to cosmetics
Anxiety
Depression
Asthma/cough
Difficulty keeping up with peers
Postprandial early fullness

The cardiopulmonary effects of PE cannot be precisely assessed by the depth of the defect alone [30]. A wide, flat PE deformity may cause cardiac compression despite appearing mild. A leftward shift of the heart with a murmur on cardiac auscultation can often be heard; the shift can be caused by distortion and compression of the heart and valves [40, 41].

Thoracic Imaging

Initial evaluation should include imaging, specifically magnetic resonance imaging (MRI) or non-contrast computed tomography (CT), which allows for visualization of the deformity revealing cardiac compression or displacement, if present, and the presence of atelectasis or tracheobronchial compression [13, 31]. The scan should be performed with both inspiratory and expiratory phases because the severity of the defect may substantially worsen when a patient exhales [42]. Imaging is used to calculate the index of severity at the lowest level of the pectus deformity and can be assessed using the Haller index or the correction index (Fig. 3a, b). If the Haller index is greater than 3.25 (normal 2.5–2.7) or the correction index score is more than 10%, it is considered severe [43, 44•, 45].

Electrocardiogram

A 12-lead electrocardiogram should be completed in all patients. Right bundle branch block and electrical aberrancies read as atrial and ventricular hypertrophy can be commonly seen in patients with PE [46]. An echocardiogram is usually necessary to properly assess chamber

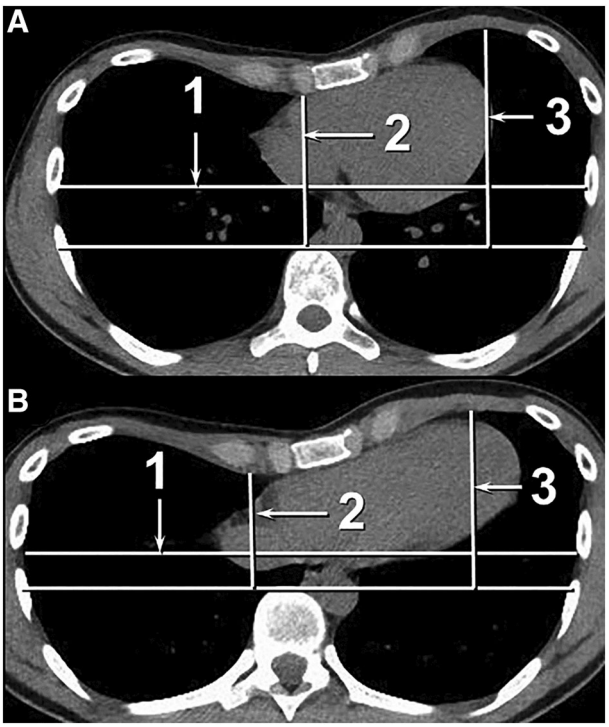

Fig. 3 Unenhanced inspiratory (**a**) and expiratory (**b**) thoracic CT obtained for pectus excavatum shows measurement of the maximal transverse thoracic diameter (*1*), and the minimum antero-posterior thoracic dimension (*2*). The pectus, or Haller Index is given by (*1*)/(*2*). The correction index is calculated as follows: (*3*)–(*2*)/(*3*) × 100%. On the inspiratory study, the maximum transverse thoracic diameter (*1*) was 258.6 mm and the minimum antero-posterior thoracic dimension (2) was 76.1, yielding a pectus (Haller) index of 3.4. The value for (*3*) was 108.6 mm, yielding a correction index of 30%. On inspiratory imaging, the maximum transverse thoracic diameter (*1*) was 253.2 mm and the minimum antero-posterior thoracic dimension (*2*) was 52 mm, yielding a pectus (Haller) index of 4.9. The value for (*3*) was, 79.9 mm, yielding a correction index of 35%

dimensions. Sinus arrhythmias and premature ventricular beats are also reported [47].

Echocardiogram

In order to rule out anatomic abnormalities an echocardiogram should be completed. Transthoracic imaging may be limited in patients with severe deformities. Echocardiogram is critical for patients who are suspected of having Marfan familial syndrome to assess aortic dimensions and mitral valve function [10]. Anatomical abnormalities in the mitral valve leaflets and right ventricular morphologic changes, including a rounded apex, trabecular hypertrophy, moderator band structural changes, and ventricular wall sacculations, are also reported [41]. Transesophageal echocardiogram (TEE) can better assess the right side of the heart in patients with PE as the deformity can obscure normal transthoracic windows. Right-sided heart

compression can be significant with associated diastolic dysfunction and decreased outflow [12•, 25, 30, 41]. The right atrium and outflow tract can be compressed, reducing atrial filling and consequently decreasing the ejection fraction. The right ventricle outflow can be notably increased after surgical correction of the PE defect and result in an increase in both right and left ventricular output [12•, 25].

Laboratory Analysis

A basic metabolic profile, complete blood cell count, and lactate dehydrogenase [LDH] level should be completed. In patients with PE, LDH can be elevated and will decrease after correction [48]. This may be due to internal organ compression; however, it is a nonspecific marker. Any additional laboratory analyses based on patient comorbidities should be completed at the physician's discretion.

Cardiopulmonary VO_2 Exercise Testing

Cardiopulmonary exercise testing (CPET) is used to help quantify the degree to which PE affects a patient's ability to exercise and consume oxygen. Cardiac limitations due to PE compressing the heart can be demonstrated by an abnormally low peak maximal anaerobic VO_2 during exercise testing [20•]. Exercise ability and peak VO_2 have been shown to be statistically improved after repair of PE [20•, 49].

Pulmonary Function Tests

Pulmonary functions may not be substantially abnormal in patients with PE; however, static studies reported the median percent predicted value lower than normal at 88% for FVC and 83% for both FEV1 and $FEF_{25–75}$ [50]. In severe cases, compression of the sternum and decreased thoracic volume can cause a decrease in forced vital capacity (FVC), tidal volume, and mixed VO_2 saturation [13, 31]. Additionally, one study found that most patients had a normal forced expiratory volume in 1 s/FVC ratio; this suggests that PE causes restrictive pulmonary disability [51].

Indications for Surgery

Surgery is recommended for symptomatic patients with a severe deformity defined by a Haller Index score of more than 3.25 or a correction index score of more than 10% [44•]. Evidence for cardiopulmonary disability includes decreased peak anaerobic VO_2, restrictive pulmonary disease, right-sided cardiac compression seen by echocardiogram or imaging, and left heart shift. Significant decrease in cardiac output as reported by echo in change from supine to sitting position should allow warrant medical evidence for repair.

Principles of Repair

Minimally invasive repair of pectus excavatum (MIRPE), also known as the Nuss, is the standard of care for surgical repair of PE in children [9, 10]. Initial attempts with MIRPE for adults resulted in higher complication rates as compared to the open Ravitch repair, causing some surgeons to recommend limiting the procedure to pediatric and younger adults [29, 52]. As the patient ages, the chest wall becomes more rigid; therefore, elevating the sternum and supporting the repair with substernal bars is more difficult [28, 53]. In the past, some surgeons have recommended limiting the procedure to pediatrics and adolescents; however, numerous publications have shown successful repairs utilizing a modified MIRPE approach [24••, 54••, 55]. Excellent results are obtained with an MIRPE approach in even patients over 30 years [24••, 29, 32, 33, 54••, 55–61].

Since the original Nuss procedure was introduced for pediatrics in 1998 [62], there have been modifications to the surgical technique and bar stabilization which have allowed successful extension into the adult population [24••, 56, 58••, 63–67•]. One important modification is the use of forced sternal elevation [67•]. This can decrease the force required to rotate bars and lateral stripping of the intercostal muscles [55, 67•, 68•, 69–71]. Variations of this have been reported by other authors with good results [55, 67•, 69–71]. Elevation of the sternum also improves visibility and the safety of mediastinal dissection and bar passage.

In older patients, two or more bars are frequently utilized by a number of experienced surgeons [24••, 54••, 66, 72••, 73]. Multiple bars can better distribute the increased pressure of the chest wall. Distributing the pressure of a more rigid chest wall helps to decrease the risk of bar rotation and malposition [66, 72••, 73]. Pilegaard [54••] reported 70% of patients over 30 years required two or more bars. In our own practice, we utilized two or more bars in 99% of patients over age 18 [24••], and 40% of patients over 30 years required three bars for complete correction. Others reported a decreased risk of migration and reoperation when multiple bars were utilized [66, 72••, 73]. In a study of PE repair in late adolescent and adult patients, single-bar repairs required reoperation for bar rotation or incomplete correction in 11.5% of patients, while none of those who had a double-bar repair required reoperation. Several authors report on the use of shorter

bars, which may lower the risk of bar rotation [74, 75]. Bars that were too long were considered a risk factor for procedure failure and need of revision by one study [76].

Bar fixation continues to be the biggest challenge in adult patients and in earlier reports, bar displacement rates were as high as 20% [77, 78]. Medial fixation with a hinge reinforcement plate [79], medially placed stabilizers [64], bilateral distal stabilizers [68•, 80] and multipoint fixation with FiberWire [24••, 66] are successful methods for bar stabilization. These and a more recently published Bridge Technique [81] have all been reported as successful methods for bar fixation in adult patients [24••, 56, 58••, 63–65, 68•, 80, 82].

Hybrid Procedure: Concurrent Osteotomy and Chondroplasty

Open osteotomy or chondroplasty is required to achieve repair in some adult patients. Complex combined deformities, extremely calcified chest walls and patients with severe asymmetry may be better served with an open repair. Additionally, older patients are reported to be more likely to require osteotomy or cartilage resection [24••, 83–85]. At our institution, over 88% of the patients ≥30 years were successfully repaired with MIRPE; however, some did require an open resection for fracture or osteotomy [24••]. This was a significantly higher percentage than in adults 18–29 years (11.3 vs 3.5%). Scoring of deformed cartilages may also reduce postoperative pain [86]. A hybrid procedure incorporating both principals of osteotomy cuts and pectus support bars is our procedure of choice for more complex deformities and in adults when sternal elevation fails [24••].

Surgical Procedure at Our Institution

MIRPE Technique

The patient is positioned supine with longitudinal gel rolls parallel to spine with the arms tucked. Patients receive antibiotic prophylaxis with intravenous cefazolin prior to the procedure. After general anesthesia induction, a double-lumen endotracheal tube is placed for single lung ventilation. TEE probe is placed to evaluate pre- and postoperative cardiac compression, function, and anatomy.

Bilateral 3-cm incisions are made at the pectoral borders, and submuscular pockets are developed. Next, through the right incision a thoracoscopic port is placed and carbon dioxide insufflation started; another 5-mm camera port is inserted on the right side superior to diaphragm under direct visualization. Forced sternal elevation is attempted, and if successful, MIRPE is completed. At the defect center, the bone clamp (Lewin Perforating Forceps [V. Mueller NL6960], CareFusion, Inc, San Diego, California) is placed into the anterior table of the sternum and attached to a Rultract retractor (Rultract Inc, Cleveland, OH) on the left and the sternum elevated (Fig. 4) [67•].

Dissection across the mediastinum is completed using a blunt tipped instrument and electrocautery. The Lorenz dissector (Zimmer Biomet, Jacksonville, FL) is introduced into the superior interspace of defect through the right interspace and brought out through the contralateral interspace. Next, a #5 FiberWire® (Arthrex, Inc, Naples, FL) is attached to the end of the dissector and used as a guide for bar placement. Bars are custom bent and sized to extend 2–3 cm beyond the anterior axillary line. A second bar is placed one or two interspaces below the superior bar. If there is an inferior residual defect following placement of the second bar, a third bar may be required (Fig. 5). The bars are rotated into position with the sternum elevated to minimize intercostal rotational forces.

If lateral stripping of the intercostal muscle occurs in the interspace of the bar, figure-of-eight FiberWire incorporating the rib above and below the interspace is used to reinforce the interspace and prevent later displacement of the bar (Fig. 6). FiberWire is used for bilateral circumferential fixation of the bars [63] in at least three sites as well as with medial fixation thru a sternal stitch. The Rultract Retractor and perforating bone clamp are removed to secure bars to the sternum. Under thoracoscopic view, a drill is used to make a hole in the sternum above and below the bar. A fascial passer is placed through the hole to retrieve the FiberWire, from inside the chest and brought out so it encircles both the sternum and bar. This is tied on the anterior chest wall under the skin.

TEE is again performed to confirm no residual compression or evidence of pericardial fluid. Marcaine 0.25% is injected into the intercostal spaces and incisions, and a 16-French chest tube is placed through the inferior port

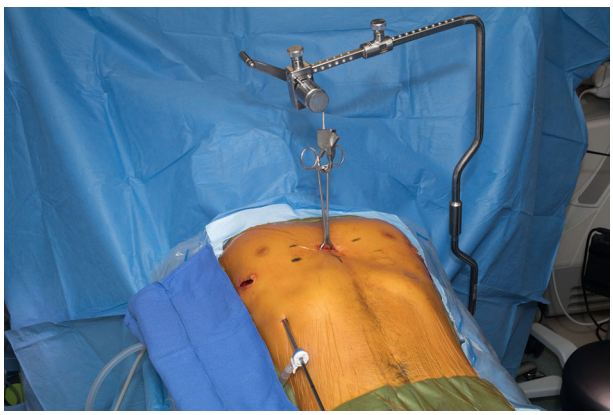

Fig. 4 Rultract elevator is attached to *left side* of table at the axilla and is attached to bone clamp to elevate the sternum

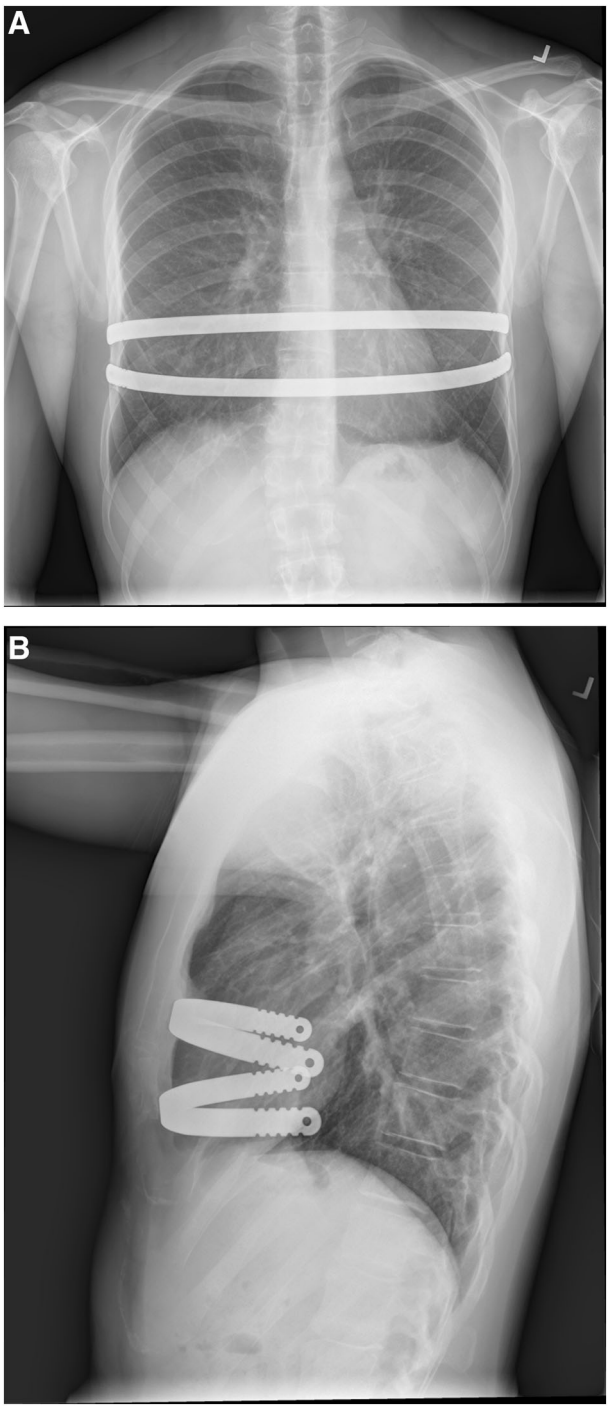

Fig. 5 **a** Antero-posterior chest X-ray with *two bars*. **b** Lateral chest x-ray

incision. Variable flow subcutaneous continuous flow catheters are placed along each lateral chest wall (7.5 inch On-Q Pain Relief System with a Select-A-Flow Variable Rate Controller, Halyard Health, Inc, Irvine, CA) (Fig. 7). Bars are recommended to remain for a minimum of three years in adults.

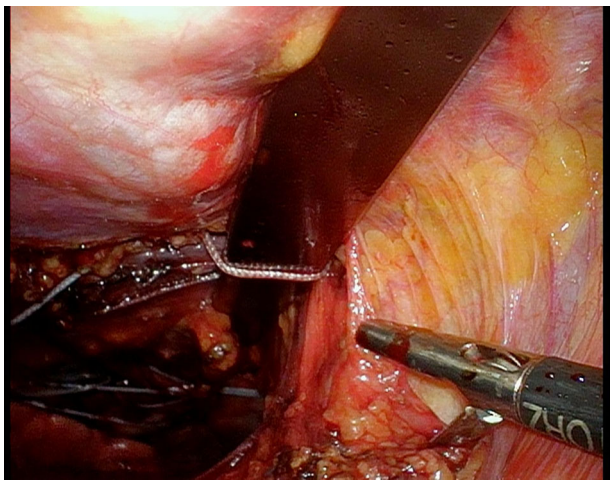

Fig. 6 Figure-of-eight FiberWire incorporating the rib above and below the interspace to reinforce the interspace and prevent displacement of the bar

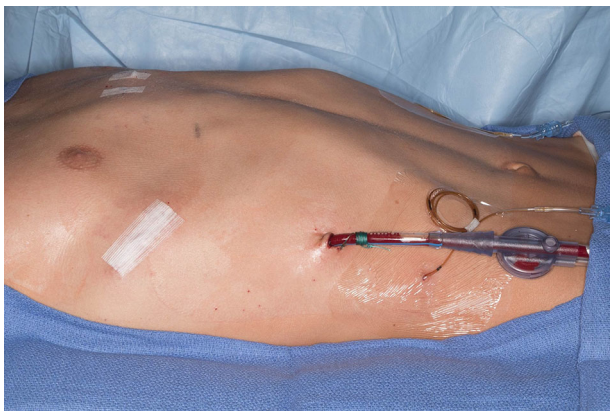

Fig. 7 7.5 inch On-Q Pain Relief System with a Select-A-Flow Variable Rate Controller (Halyard Health, Inc, Irvine, CA) are tunneled subcutaneously laterally on each side of the chest. They are left in place and refilled on patient discharge for a total of 5 days

Technique for Failure to Elevate the Sternum

If the defect failed to elevate with the use of forced sternal elevation, then a hybrid approach for repair is required. A limited midline incision is made and the pectoral muscle elevated at the affected site. Resection of a small piece of cartilage with careful preservation of perichondrium is performed to release fixed sites until the defect is elevated. Osteotomy of the sternum is performed only if elevation is otherwise unable to be achieved [87, 88]. The support bars are then placed and secured thoracoscopically as described previously for MIRPE [63]. Approximation and stabilization of resected cartilage to the sternum and sternal osteotomy sites is completed with either FiberWire or titanium plating.

Perioperative

- Gabapentin, 600mg, PO
- Celecoxib, 400mg, PO
- Acetaminophen, 1000mg, PO
- Dexamethasone, 8mg, IV
- Methadone, 0.2-0.35 mg/kg, IV
- General anesthesia induced: single dose of IV fentanyl, 0.5-2 mcg/kg; lidocaine, 1-2 mg/kg; and propofol, 2-4 mg/kg
- Subcutaneous On-Q catheters: ropivacaine, 0.2%, at 7 mL/h
- Intercostal block 1cc/kg/0.25 % Marcaine with 10 mg dexamethasone (by patient weight up to 60mg)

↓

Postoperative day 0

- PCA
  - IV hydromorphone, 0.2 mg, at 8-min demand interval dosing with a 4.8 mg lockout at 4 h (increase in on-demand dosing up to 0.4 mg allowed if necessary for a pain level consistently above 4)
  - Begin in recovery room and maintain with demand dosing only
- Ketorolac, 30 mg, IV, every 8 h
- Gabapentin, 300 mg, PO, every 8 h
- Acetaminophen, 1000 mg, PO every 6 h
- Diazepam 5mg, IV, every 6 h

↓

Postoperative day 1 -- discharge medications

- PCA discontinued the morning of postoperative day 1
- Morphine ER initiated, 10-20 mg, oral, every 12 h Oxycodone, 5 10 mg, PO, every 3-4 h as needed
- Ketorolac discontinued; ibuprofen initiated, 600 mg, PO every 8 h
- Gabapentin, 300 mg, PO, every 8 h
- Acetaminophen, 1000 mg, PO, every 6 h
- Diazepam 5 mg, PO, every 8 h PRN

Fig. 8 Standardized protocol pain medications. *PO* oral, *IV* intravenous, *PCA* patient controlled analgesia, *ER* extended release, *PRN* as needed

Postoperative Care

Postoperative pain remains a concern for adults undergoing Nuss [89]. Several authors have discussed various analgesic regimens to achieve adequate pain control and ensure the patient comfort immediately after surgery [90]. The use of thoracic epidurals, intravenous on- demand patient-controlled anesthesia, subcutaneous continuous flow catheters and local paravertebral blocks have all been reported with successful pain control in the immediate postoperative period [91–95]. Tunneled, anesthetic-infiltrating catheters are being used increasingly after thoracic procedures in lieu of thoracic epidural analgesia (TEA) with studies showing earlier hospital discharge for patients who receive this type of analgesia [92, 93, 96]. In our experience, a standardized polypharmacy protocol achieved good pain control in addition to delivery of local anesthesia by subcutaneous continuous flow catheters [24••, 91, 97]. Figure 8 reviews our protocol for preoperative and postoperative pain control.

Outcomes in Adults After PE Repair

There are several publications documenting improved symptoms and quality of life (QOL) in adult patients after Nuss repair [57, 98]. Krasopoulos et al. [57] proposed the two-step Nuss Questionnaire modified for Adults (NQ-mA) and the Single Step Questionnaire (SSQ). A significant improvement in self-esteem, social functioning and a high satisfaction following Nuss was found. Pain was a significant concern in the immediate postoperative period, which decreased significantly after several weeks; however, all patients were off analgesics 4–5 months after surgery.

Other surgeons have utilized this modified survey for postoperative assessment [35•, 98]. Hoksch et al. [98] reported a prospective study on the long-term results of Nuss in adults using NQ-mA & SSQ. This study included a larger adult cohort ($n = 129$) initially, but is limited by 19 patients observed for >10 years after surgery. The results obtained after surgery were in the follow-up period of 3, 12 and 36 months and showed high levels in QOL improvement 88, 89% and increased to 92%, respectively. Even > 10 years, continued improvement in QOL was confirmed in 57.9% of patients, and Nuss was recommended by 94% of patients.

Hanna et al. [34] investigated the mid-term results in young adults undergoing Nuss repair and evaluated them using the single-step QOL survey. They reported an improvement in both self-esteem and social life. There was a 92% subjective improvement in the chest wall appearance.

Conclusions

Adult patients may present with progressive symptoms from their PE deformity. A thorough workup is indicated and repair recommended if cardiopulmonary deficit is identified. In adults requiring surgery, the use of a modified Nuss for pectus repair can be effective with low complication rates. Techniques for sternal elevation can improve success in adults with a more rigid chest wall. In addition, multiple bars and improved methods for bar fixation have reduced complications and bar rotation. Overall, patients report high satisfaction and improvement in symptoms following repair of their PE.

Compliance with Ethics Guidelines

Conflict of Interest The authors declare no conflicts of interest relevant to this manuscript.

Human and Animal Rights and Informed Consent This article does not contain any studies with human or animal subjects performed by any of the authors.

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