



## Pectus Excavatum

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### Introduction and Epidemiology

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Pectus excavatum (PE), or “funnel chest”, is a malformation of the chest wall characterized by a sternal depression typically beginning over the mid-portion of the manubrium and progressing inward through the xiphoid process. Mayer Pectus excavatum is the most common congenital malformation of the chest wall (90%) with an incidence of 1 in every 400 to 1000 live births, and, for reasons not well understood, a male-to-female ratio of 4:1. Mayer, Aloi Depending on the severity of the defect, deviations of thoracic organs and spine deformities are known. Brochhausen PE can lead to cardiopulmonary dysfunction as well as psychological distress which can require psychological therapy.

The majority of cases of pectus excavatum are sporadic, but up to 40% of pectus excavatum patients report a family history of major or minor chest wall malformations, suggesting a genetic etiology for this portion of patients. Pectus excavatum has been found in twins and occurring in subsequent generations of some families. Members of these families have been demonstrated to be carriers of connective tissue disorders, such as Marfan’s syndrome (5–8%), Poland’s Syndrome (5.4%), Ehlers–Danlos Syndrome (3%) or Sprengle’s Deformity (0.6%). Aloi The association of pectus excavatum with connective tissue disorders supports the hypothesis that pectus excavatum occurs due to an intrinsic abnormality of the costochondral cartilage. Two percent of patients with pectus excavatum also have comorbid congenital heart disease and 15-20% has scoliosis. Pectus excavatum also appears as a component of several rare syndromes, many involving mental retardation or neurological disorders such as the King-Denborough Syndrome, spinal muscular atrophy, Noonan Syndrome, Turner Syndrome, and multiple endocrine neoplasia type 2b. Aloi

### Pathophysiology

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The precise pathophysiology of pectus excavatum is unknown, but it is thought that a defect in the metabolism of sternocostal cartilage causes a defect in the cartilage structure and growth. Variable cellularity, matrix disorganization, and premature aging of the sternocostal cartilage are seen in pectus excavatum. Brochhausen Ultrastructural and biochemical studies of the content of trace elements in the costal cartilage from PE

patients demonstrated decreased levels of zinc and increased levels of magnesium and calcium, supporting the hypothesis that low levels of dietary zinc results in a lower metabolic activity of chondrocytes. Brochhausen

## Presentation

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The sternum appears depressed with the abnormal angulation often apparent beginning with ribs two through seven. The lower-most ribs may protrude laterally, giving younger patients a “pot belly” appearance. Dean Other features commonly associated with PE include: long arms, legs, and digits, high-arched palate, hypermobility of joints, flat feet, childhood myopia, poor healing, and easy bruising. Abnormal posture with dorsal lordosis, and or thoracic kyphosis is frequently observed in conjunction with PE. Furthermore, some patients may develop scoliosis. Dean Infants and young children rarely have symptoms, but many will develop symptoms of cardiopulmonary dysfunction, most often noted as exercise intolerance, as they enter into adolescence. Frantz Symptoms worsen at this age due to rapid skeletal growth during puberty. The most consistently expressed symptoms are dyspnea with exercise and loss of endurance. Jaroszewski Of those exhibiting cardiac symptoms; chest pain, tachycardia, palpitations and heart arrhythmias are the most common complaints. Dean

In addition to any somatic complaints, the majority of patients will experience psychosocial distress due to their abnormal appearance. One study found that sixty-eight percent of female patients and 40% of male patients reported that they were often distressed by the appearance of their chests. Dean PE is an under-recognized cause of psychosocial stress and contributing factor to mental illness in affected children and adolescents. Patients may become extremely self-conscious, frequently withdraw from social and sports activities, develop body image difficulties, interpersonal difficulties, and can become depressed and even suicidal. Nuss

## Prognosis without surgical intervention

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Foregoing surgical correction of the deformity carries a risk of worsening of the defect, particularly as the child progresses through adolescence, and a high risk of a negative psychosocial impact on the adolescent, no matter how severe the defect. About one third of cases of PE present in infancy. Mayer Pectus excavatum often does not become clinically apparent until puberty. A delay in onset may be observed when PE is a component of Marfan syndrome. Dean Spontaneous regression of PE in infancy is rare. Mayer The frequency of spontaneous improvement decreases further after one year of age, and no spontaneous improvement can be expected after six years of age. Mayer After 12 years of age, the PE deformity worsens in one-third of patients during the adolescent growth spurt, and remains the same in two-thirds. Mayer There are no reliable markers to predict progression. Mayer As PE worsens, simple symmetrical lesions may progress to more complex asymmetric deformities. Mayer No debilitating disabilities or deaths have been attributed to PE in children or young adults. Mayer In adults, complaints of exercise intolerance are common; no deaths have been attributed

to the isolated deformity. Mayer

## Physical findings and operative planning

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Morphological classification of the chest wall has practical implications as certain variants are known to have better surgical outcomes with the repair utilizing the traditional Nuss procedure, a modified Nuss procedure, or an open repair. "The ideal chest configurations for the minimally invasive repair are the diffuse saucer shape, localized cup shape, and symmetric funnel shape.<sup>9</sup> Patients who have very steep cup shaped depressions and those with severe deep, asymmetric, Grand Canyon type depressions are more of a challenge and often require 2 bars. Park and colleagues<sup>6</sup> and other researchers<sup>7</sup> have suggested using an asymmetric bar in these patients. In patients in whom the depression mostly involves the upper chest, care needs to be taken not to place the bar too high because such placement will interfere with the axilla and its vital structures. Patients who have mixed excavatum and carinatum deformities may have residual protrusion of the carinatum after bar placement, especially if there is severe sternal torsion. Older patients have a higher incidence of sternal torsion and mixed deformities, which may be a good reason to perform the repair before puberty.<sup>17,67,68</sup> Patients who have Currarino-Silverman Syndrome or Pouter Pigeon deformity with anterior displacement of the manubrium and posterior displacement of the gladiolus develop increased protrusion of the manubrium when the gladiolus is elevated with a substernal bar. Therefore, the minimally invasive procedure is not recommended in this category of patients."Nuss

The severity of pectus excavatum can be measured quantitatively with the pectus index (Haller index), which is the ratio of the internal transverse diameter of the thorax to the diameter measured from the vertebrae to the most depressed portion of the sternum as measured by computed tomography (CT).Dean Cardiopulmonary dysfunction increases with a rising Haller index, and patients with an index over 4.9 develop significant cardiac and pulmonary dysfunction.Swanson In their study of 33 patients selected for surgical pectus repair and 19 age matched control patients, Haller et al. found that all the surgical cases but none of the controls had a HI exceeding 3.25, suggesting HI as an objective criterion for pectus excavatum repair.Swanson The Haller Index is not useful for patients with the eccentric or barrel-chest type deformities as they will have a PSI <3.2, even though their defect is considerable and often warrants correction.Krasapoulos In addition, the high variability of other features such as the position of the apex of the depression, depth and extent of the depression, the ratio of the sagittal to lateral diameters of the thorax, and the protrusion of the costal arches makes the deformity still difficult to quantify.lida

The apex of the depression may be in the midline but is commonly offset to the right, creating an asymmetrical funnel[24, 33]Dean Park et al. described a thorough morphologic classification system that designates the deformity according to its symmetry, depth, and width as measured by computed tomography. For the Nuss

procedure (see below), surgical outcomes were improved when with the bars used for fixation were shaped pre-operatively in order to control the amount of pressure applied to each area of the chest wall.

A cardiac evaluation is indicated as a significant number of patients have findings of right atrial and ventricular compression leading to a diminished stroke volume, mitral valve prolapse, and rhythm abnormalities. Nuss The sternal depression displaces the heart toward the left, such that the point of maximal impulse is often palpable near the left mid-axillary line. Dean "On auscultation, a split second heart sound (S2) is heard in nearly all patients, and systolic murmur is heard in about half of the patients due to compression of the right ventricle by the chest wall [17, 22]. Anteroposterior chest radiograph may reveal an obscured right cardiac shadow in patients with moderate to severe deformity and an increased cardiothoracic ratio in patients with mild PE. These effects on the heart cause characteristic electrocardiographic findings, such as negative P wave in lead VI as well as negative T waves in V1, V2 or V4. Incomplete right bundle branch block is also common [17, 22, 37]. In addition, mitral valve prolapse has been reported in 15% of PE patients [25]." Dean

Pectus repair has been successfully performed on patients from age 1 year to older than 50 years Nuss, but most authors agree that the optimal age for pectus repair is at least prior to puberty. The minimum age at which each procedure is indicated is a subject of debate. In 1989, regarding repair with the Ravitch technique, Haller et al. suggested an ideal age of 4–6 years on the basis that these patients had reached a level of maturity sufficient to cope with hospitalization and to refrain from "roughhouse" activities postoperatively. They found that operating earlier than 4 years of age did not improve outcomes. In 1996, they revised their recommendation to at least 6 to 8 years of age after 12 of their patients developed severe cardiorespiratory compromise secondary to restriction of chest wall growth, now commonly referred to as acquired asphyxiating chondrodystrophy. This disruption is thought to be attributable to damage of growth centers in the developing children's ribs. Haller, Dean, Chen

Park et al. sought to establish an accepted minimum age for repair using the minimally invasive Nuss procedure. Their retrospective chart review of the Nuss procedure performed in children as young as three years old showed that these youngest patients did not suffer any impairment of chest wall growth. Nuss et al. state that the ideal age for the minimally invasive procedure is just before puberty because at that age the chest is still malleable, the support bar is in place during the pubertal growth spurt, the recovery time is short, the incidence of recurrence is low, and relief of the cardiac and pulmonary compression at an early age allows these organs to develop fully.

Although many patients with PE have a perceived limitation of exercise capacity, investigations of exercise ability have yielded mixed results. Nuss Some propose that increasing severity of the pectus depression is directly related to increasing loss of physical work capacity. Nuss Others found that many patients with subjective complaints of exercise intolerance had no measurable deficits of cardiopulmonary performance.

Studies on the effect of pectus excavatum on pulmonary function alone have also yielded mixed results. There is not a direct correlation between the severity of the defect and pulmonary function as measured by spirometry. One study found that pulmonary function test was abnormal in only 67% of children with PE and of those, reduced vital capacity was the most common finding. Bohosiewicz While pulmonary function and exercise testing haven't been shown to be reliable tools for preoperative assessment, the evaluations still provide valuable data for future research.

The following abbreviated guideline is helpful for selection of patients for surgical correction: "Patients are selected for surgical correction if they demonstrate two or more of the following criteria: symptoms; history of progression of the deformity; paradoxical movement of the chest wall with deep inspiration; a chest CT scan with Haller index greater than 3.25; cardiac compression, displacement, or pulmonary compression identified on echocardiogram or CT; abnormal pulmonary function studies showing significant restrictive disease; mitral valve prolapse, bundle branch block or other cardiac pathology secondary compression of the heart; history of failed previous repair(s); and significant body image disturbance." Kelly, Nuss

## Surgical Technique:

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### Introduction

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The two procedures most often performed for the correction of pectus excavatum are the open method, the "highly-modified Ravitch procedure," and the minimally invasive "Nuss procedure," first described by Dr. Ravitch in 1949 and Dr. Donald Nuss in 1998, respectively.

### Nuss Technique

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The Nuss procedure is considered minimally invasive because it does not require sternotomy or resection of costal cartilages as is necessary in the Ravitch procedure, but instead only requires a few lateral incisions. Briefly, prior to the insertion of the bar, a transthoracic, substernal tunnel travelling anterior to the heart is created using an introducer under thoracoscopic guidance. Frantz Then, the arc-shaped bar is inserted with the convexity posterior and then rotated such that the convexity is anterior, displacing the sternum anteriorly. The bar is often fixed using stabilizers or sutures to maintain its position for several years until the cartilage has remodeled and the bar can be removed. The ideal material for the bar is surgical steel rather than titanium because the steel bars have elastic recoil in addition to adequate strength to support the chest in the corrected position if the child sustains trauma. Nuss The bar should be long enough to accommodate growth for two years. Nuss

The standard approach to the minimally invasive technique, including description of patient positioning, thoracoscopic technique, skin incisions, tunneling, sternal elevation, and bar stabilization have been described in detail elsewhere. Nuss

Patients with certain comorbidities or anatomic variants will require special consideration. Older patients, due to the larger volume of their thoracic cage, often require two support bars. Those with Marfan's syndrome or other connective tissue disorders have softer bones and have better outcomes using two bars to distribute the pressures over a wider area. Nuss Those with an allergy to nickel may receive titanium bars to avoid this complication.

## Modifications to the standard Nuss procedure

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Displacement of the support bar has been a common complication of the Nuss procedure. Without stabilizers, bar displacement was found to occur in up to 15% of cases. Nuss Many modifications have been described involving methods of fixation with stabilizers to prevent bar dislocation. Clark The development of stabilizers has decreased the incidence of bar displacement, but the different materials, placement, and design of each type of stabilizer present unique challenges.

Switching from rectangular stabilizers to triangular stabilizers and the placement of a circum-costal wire for additional fixation has been reported to reduce the rate of bar displacement to 5% in one study. Nuss A pericostal suture can be placed around one costal arch, often through the right lateral thoracic incision, and a stabilizer placed on the left side. Hebra

A disadvantage of metal stabilizers is that they often cause chronic pain that requires a second intervention for removal. Pilegaard An alternative, slowly absorbable stabilizer made of L-lactic and glycolic acids (Lactosorb®) was introduced in 2007. In one study, of the 85 patients with absorbable stabilizers placed, three stabilizers broke within 6 weeks of surgery (3.5%) and another three absorbable stabilizers (3.5%) dislocated within 8 weeks of surgery without any connection to physical activity. Pilegaard

There is much concern for increased infections with use of stabilizers of any material. However, it was found that while the stabilizers did increase the incidence of wound infections, the infections were initially aseptic seromas with dermatitis due to pressure damage that then became infected after surgical drainage. Watanabe

The standard technique has been to place stabilizers at the bar edges, but Milanez et al. proposed that placement of two stabilizers in the intermediate region between the bar edges and the mid-point, close to the entry and exit of the bar in the intercostal space, posterior to the muscles, but not close to the cartilage, would be less likely to become displaced. They used stabilizers with central grooves on the posterior surface that allowed for the stabilizer to slide easily, regardless of the bar's curvature. The disadvantage of this technique is that it requires the additional dissection of the pectoralis major muscles medially to allow the stabilizers to be placed posterior to them. Milanez et al. used this technique in two patients, without any additional morbidity or complication incurred.

The edges of the bar are serrated, and can cause tissue damage and hemorrhage when

it is removed, particularly if the patient is not maintained in a symmetrical position throughout removal. Nuss, Milanez et al. have introduced a technique in which the serrated edges of the bar are enveloped in a protective film to protect the surrounding tissue during removal. This technique was used for one patient, without any additional morbidity or complication incurred.

To further minimize scarring, the Nuss procedure can be performed using only a right lateral thoracic incision with results comparable in safety and efficacy to the standard Nuss technique. Clark Through a single right lateral thoracic incision and under thoracoscopic guidance, a dissection across the chest in a subcutaneous, pre-sternal, and pre-muscular plane forms a tunnel into which the bar can be placed in the standard position. No statistically significant differences between the study groups in any of the patient, operative, or postoperative care parameters were reported. Clark

## Outcomes of the Nuss Procedure

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Surgical repair of pectus excavatum with the Nuss procedure has been shown to improve parameters of cardiovascular function such as cardiac stroke volume, cardiac output, forced expiratory volume in the first second of expiration (FEV1), total lung capacity (TLC), TLC (percentage expected), diffusing lung capacity, maximum oxygen consumption (VO<sub>2</sub>max), respiratory quotient, and O<sub>2</sub> pulse (percentage predicted). Chen A study on the end-diastolic right ventricular dimensions and left ventricular ejection fraction of adults using intraoperative transesophageal echocardiography immediately before and after surgical correction of pectus showed that both parameters immediately and significantly increased after surgical correction. Krueger

There is a modest preoperative reduction in vital capacity and total lung capacity, both of which improve after surgical correction. Chen FEV1, the amount of air that can be forcefully exhaled in one second, was improved postoperatively. This indicates that the repaired chest wall allows the extrinsic musculature and diaphragm to facilitate more efficient breathing. The Ravitch and Nuss procedures effect equal improvement in pulmonary function within 1 year, but greater improvements were seen for Nuss patients after bar removal. Chen

Others found that postoperative Nuss procedure patients had no change in lung function

“Postoperatively, patients who have undergone the Nuss procedure typically experience a temporary decrease in forced vital capacity, which resolves with removal of the implanted bar [6]. The likely explanation is that the bar restricts maximal thoracic expansion. In support of this, a study by Castellani et al. [6] showed no significant differences in preoperative and postoperative lung function.” Dean

The measured improvements in cardiopulmonary function may be confounded by the post-operative cardiorespiratory rehabilitation that patients receive, improved optimism toward exercise goals, and increased social integration.

The Single Step Questionnaire (SSQ), is a questionnaire designed to assess the impact of surgical correction of PE on patient's quality of life and psychosocial well-being. Administration of the questionnaire to forty patients at 6 months after MIRPE with the bar in place and again to the patients and their parents at a mean of 23 months after bar removal demonstrated a significant improvement in parameters linked to self-perception, self-esteem and social behavior. Metzelder

## Nuss Complications

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Common complications of the Nuss procedure include pneumothorax, pleural effusion, pain, wound infection, and bar displacement. Vegunta Less common complications include cardiac perforation, aortic perforation, pericarditis, brachial plexus injury, diaphragmatic perforation, and thoracic outlet syndrome. Vegunta The use of thoracoscopy has significantly decreased the incidence of cardiac complications, but perforations have occurred despite this protective measure. Bouchard Left chest thoracoscopy with blunt dissections provides improved mediastinal visualization and a decreased risk of cardiac injury, except in those with severe deformities and a leftward displaced heart. Bouchard The support bar exerts a force that ultimately is absorbed by the spine. If the malformation is asymmetric, the spine may bend from the side of the chest with greater concavity to the side with less concavity, resulting in scoliosis. Interestingly, this remodeling may be advantageous if the side of spinal bowing coincides with the side of anterior wall concavity because the spinal bowing may be corrected by that counterforce. Dean

## Ravitch Technique

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"In 1949, Ravitch described a technique (open technique) involving dissection of the pectoralis major and rectus abdominis, followed by subperichondrial resection of all deformed cartilages, xiphoid excision, and sternal osteotomy with anterior fixation of the sternum." Dean Modifications to the Ravitch technique have focused on experimentation with different materials for posterior support of the sternum.

Termed the "Robicsek technique," a modification of the Ravitch procedure involving the use of DualMesh 2-mm Gore-Tex to provide posterior support of the sternum has been reported on with great results by several authors. Kotoulas, Spartalis Spartalis et al. report the results of a 17 year old patient who underwent this modified Robicsek technique. Their technique is described as follows: the mesh is placed under the sternum and anchored to the lateral tips of all divided costal cartilages to stabilize the chest wall. The previously detached edges of pectoralis major and rectus abdominis muscles were sutured over the anterior sternal surface. The pectoral muscle flaps were secured to the midline while being advanced to provide coverage of the entire sternum. The rectus abdominis muscle flap was joined to the pectoral muscle flaps. The patient had no postoperative complications and had excellent cosmetic results. A high resolution three-dimensional reconstructed computed tomographic scan taken seven years post-operatively confirmed that the mesh was still stably anchored and supporting the

sternum in its corrected position. Two main advantages of the Gore-Tex mesh are that it is very resistant to infections and it has two different surfaces: one smooth for minimal tissue attachment and the other textured for tissue ingrowth. Kotoulas The textured surface is positioned adjacent to the posterior sternum and causes creation of adhesions, while the smooth surface faces the pericardium and does not cause adhesions to form. Thus, the authors propose that the material may stay in place permanently whereas at the same time it can be detached easily from the pericardium in the case of an urgent sternotomy. Kotoulas Kotoulas et al., with 10 years experience using the 2-mm Gore-Tex mesh support in 21 young adult patients, reported "excellent cosmetic results, no cases of infections, no cases of recurrence, and no limitations in activity."

## Comparison of Nuss and Ravitch Procedures

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Advantages of the Nuss procedure are that it avoids extensive dissection, cartilage resection and osteotomy, which are integral components of the Ravitch technique. Dean It requires a shorter operating time, results in only minimal blood loss (in the 10-30 mL range), and allows for an earlier return to full activity. Nuss It provides superior cosmetic results, which is often an important consideration for patients as cosmesis is often a primary motive for seeking correction. Krasopoulos

Compared to pre-adolescents, adult patients undergoing the Nuss procedure often experienced more intense pain postoperatively, required prolonged postoperative care, and had a higher incidence of bar displacement and recurrence; all of which may be attributable to the increased rigidity of the chest wall in these older patients. de Souza Coelho Outcomes for the Nuss procedure performed on adults were better if two bars and/or lateral stabilizers were used. Dean Patients with asymmetric malformations who were corrected using the Nuss procedure were more likely to be over corrected and develop postoperative pectus carinatum than those with asymmetric malformations who underwent the Ravitch procedure. de Souza Coelho Thus, open repair should be considered for the older patient or the patient with an asymmetric malformation.

## Failed Repairs and Recurrence

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The recurrence rate after repair of PE is reported to be anywhere from 2% to 37%. Croitoru Recurrences are more common in those with Marfan's Syndrome, those whose repair was completed at either a very young or an advanced age, those with either too extensive or too little dissection with an open repair, premature removal of the bar in minimally invasive repairs, local infections, or an unsatisfactory primary result. Croitoru Patients with a failed repair or recurrence will often present with the same symptoms they experienced before the primary repair, such as shortness of breath, chest pain, and other similar symptoms that affect their activity level. Croitoru Associated cardiac findings such as cardiac displacement and cardiac compression were often still present. Croitoru In addition, these patients continued to perform poorly on pulmonary function tests (PFTs). Croitoru Patients with a failed Nuss procedure will usually present shortly after the

primary surgery with bar displacement will be evident on chest x-ray. Croitoru Patients with a failed Ravitch procedure or a recurrence after such may present immediately after or up to seven years postoperatively. Croitoru Those that present remote from the primary surgery are often those that had a successful repair performed before the pubertal growth spurt but the thoracic cage failed to grow appropriately during puberty. Croitoru

Croitoru, et al., reported excellent results with the minimally invasive Nuss technique for recurrent or failed pectus excavatum repair. Their study demonstrated that the Nuss procedure was superior to the open procedure for a repeat repair. The inherent extensive dissection performed during an open procedure, although technically extrapleural, can produce extensive intrathoracic adhesions. Also, most patients with a previous Ravitch repair will have poor chest wall movement and compliance because of the abnormal ossification of the anterior chest wall secondary to the previous invasive procedure. Repeating the open procedure would necessitate another violation of the perichondrium and periosteum, thus stimulating more ossification, calcification, and subsequent loss of chest wall movement and compliance. For this reason, Croitoru et al., also do not advocate wiring of the pectus bar to the ribs or sternum. In patients undergoing a second repair, "complications were slightly higher than those in primary repairs and included pneumothorax requiring chest tube (14%), hemothorax (8%), pleural effusion requiring drainage (8%), pericarditis (4%), pneumonia (4%), and wound infection (2%). There were no deaths or cardiac perforations. Initial postoperative results were excellent in 70%, good in 28%, and fair in 2%. Late complications of bar shift requiring revision occurred in 8%. Seventeen patients have had bar removals with 9 patients being more than 1 year postremoval. For the 17 patients who are postremoval, excellent results have been maintained in 8 (47%), good in 7 (41%), fair in 1 (6%), and failed in 1 (6%). There have been no recurrences postremoval." Croitoru Although postoperative cardiology evaluations or repeat CT scans were not performed, symptoms of cardiac compression and displacement were found to be relieved postoperatively. The patients who had a secondary minimally invasive procedure after either a primary Ravitch or Nuss procedure and who returned for follow-up reported resolution of their pre-operative symptoms with 85% reporting increased exercise tolerance. 82% of respondents (23/28) reported satisfaction with their results. Croitoru

## Summary

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Pectus excavatum is a common, congenital chest wall deformity in which the sternum is depressed. In severe disease, the patient may experience cardiopulmonary dysfunction, most commonly described as exercise intolerance. The minimally invasive Nuss procedure and the modified open Ravitch procedure are both safe and effective techniques for repair of pectus excavatum. The choice of which procedure to perform should be made on an individual basis. Multiple factors such as the patient's age,

comorbidities, severity of the defect, and symmetry of the chest wall should all be considered in pre-operative evaluation. Research is ongoing and surgical techniques are continuously evolving as new modifications are introduced.

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