MINI REVIEW ARTICLE

Surgical correction of recurrent pectus excavatum of an adult patient, case report, and review of literature

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Abstract
Purpose The aim of this paper is to review the literature on recurrent pectus excavatum (PE) and present our surgical approach to a complex case of recurrent PE in an adult patient at a Mexican Hospital.
Methods We present the case of an adult patient with severe and symptomatic PE, with history of a failed Nuss procedure 1 year previous our intervention, which consisted of a combination of both classic techniques, by performing an osteochondrectomy of affected cartilages and placing a titanium bar substernal and stabilizing coastal arches with secondary osteosynthesis system (Stratos ™ system, medXpert, Germany).
Results Adequate correction of thoracic silhouette and both cardiac and respiratory disorders in the 1-year follow-up was achieved as indicated by the improvement of the patient's Haller index.
Conclusion Successful surgical correction of pectus excavatum is achieved when the thoracic silhouette is restored, thus improving cardiopulmonary symptoms. As there are many different techniques available, the more minimally invasive ones are reserved for mild cases, but the treatment of complex cases as in our patient requires a combination of multiple techniques and reconstruction materials in order to achieve adequate correction of the thoracic deformity and reduce recurrence rate.

Keywords Pectus excavatum · Chest wall repair · Osteosynthesis · Reconstruction · Deformity · Recurrence

Introduction

PE is the most common thoracic abnormality. It is a deformity in the growth of the sternum that creates a sunken chest and may cause severe and symptomatic disease, which must be corrected surgically. The Ravitch procedure has been used for decades as an open, invasive technique, while the Nuss procedure is considered minimally invasive, but its main use is for young patients. Both types of techniques have similar recurrence rates and a second attempt to correct PE is often required, which becomes more challenging to solve surgically. Identification of the cause of recurrence is critical because a wide variety of hybrid techniques and materials can be used to achieve satisfactory results in a recurrent PE correction surgery.

Clinical presentation

PE comprises 75% of all thoracic wall malformations with an overall incidence of 8–100 live births, a male-to-female ratio of 2:1, and about 86% of cases are noticeable at birth. PE is considered a congenital disease that might be mild at birth, but as the patient grows PE progresses, it is associated with other musculoskeletal system abnormalities, such as scoliosis (30%), and connective tissue diseases, such as Ehlers-Danlos or Marfan syndrome (15%) [1–5].

The symptoms are proportional to the degree of the thoracic concave deformation, which range from asymptomatic and treated conservatively to severe with symptoms that might require surgery. Patients with severe defects suffer from depression, social dysfunction, and poor body image [2]. The patients' most commonly registered complaint is the unattractive physical appearance of deformity.

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Thoracic computed tomography (CT scan) is the gold standard for diagnosis confirmation and preoperative evaluation, which helps document the severity of the deformity and determine the presence of cardiac or pulmonary compression and displacement. This tool is an important aid in the objective decision-making for when and how to approach surgery [3].

Surgical considerations

Surgical correction of PE is offered to any patient that fulfills two or more of the following criteria: severe or progressive deformity who experience exercise intolerance, chest pain, or shortness of breath; Haller index (i.e., a pectus index ratio of thoracic width and height) greater than 3.25 points; spirometry that indicates either obstructive or restrictive pulmonary dysfunction; cardiac evaluation that demonstrates compression or displacement of the heart, mitral valve prolapse, arrhythmias, or murmurs; recidivating PE is also an indication for surgery [1, 2].

Considering the timing for surgery, surgical procedures and outcomes are assumed to be better in a younger patient because PE is a progressive deformity that worsens with the patient's growth, rapidly evolving from mild to severe, which complicates surgical repair [1].

Pectus excavatum repair techniques

The first interventions for PE surgery in the early 1990s consisted of chondrosternal resection, but this affected the thorax protective role for the heart and respiratory dynamics, and poor cosmetic outcomes also developed. External traction was introduced but abandoned as an adjuvant technique because it was impractical and led to infection. However, procedures without traction showed a higher recurrence rate. In 1950, internal fixation was introduced, but longer surgery time and complications, such as blood loss and failure of repair, were reported.

In 1949, Ravitch introduced a technique that involved subperichondrial resection of all deformed costal cartilages, xiphoid process excision, and sternal osteotomy with anterior fixation of the sternum. It was considered the procedure of choice for a long time with minor modifications, such as in 1958, when Welch extended the costal cartilage resection from partial to total and included mobilization of the sternum [3, 4]. In the late 1990s, Nuss introduced a minimally invasive repair of pectus excavatum (MIRPE) approach without any chondrectomy, using the younger patients' chest wall plasticity and inserting a convex steel bar under the sternum through two lateral incisions, aided by thoracoscopic direct vision for correct positioning and fixation to the chest wall laterally. The steel bar itself corrects the deformity but must be surgically removed from 2 to 4 years later [2, 3].

Both the surgeon and patient must make the decision on which surgical technique to use based on the surgical expertise of the former and once all the advantages and disadvantages of both procedures are explained to the latter [1]. The advantages of MIRPE include no anterior chest wall incision, no resection of tissue, shorter surgical time, minimal blood loss, early return to activity, and excellent long-term cosmetic results, which make this approach very appealing even for older patients [3, 5].

The recurrence rates for PE repair have been reported in 2%–37% of patients for both open and MIRPE techniques. The causes were often derived from the initial repair, but no long-term studies for PE recurrence have been performed to recognize individual risk factors.

After primary MIRPE, recurrence may present mainly due to either technical factors, such as bar displacement (i.e., lateralization, inadequate length, inadequate bar securing) or failure to correct the defect (i.e., rigid chest wall, inadequate number of bars to support chest wall). Thoracic stability is impacted by the number of bars placed, but there is no consensus for the number of bars required to achieve thoracic stability. Older patients have been reported to require two or more bars for a stable PE repair, while early removal of the pectus bar is also associated with recurrence [6–9]. Recurrence after primary Ravitch procedure and its modified alternatives is often related to incomplete excision of diseased cartilages or extensive dissection failing to preserve the perichondrium, incomplete healing or failed fusion of excised cartilage and sternum, infection and seroma, and failure to support repair or too early removal of support [10].

Once recurrence presents, a subsequent repair can be more complex due to chest wall rigidity and scar tissue from the prior intervention. These factors may be recognized as a relative contraindication for another MIRPE; therefore, open repair and stabilization are recommended for these cases [10, 11]. Indications for correction of PE recurrence are similar to the ones for primary repair, in addition to areas of nonunion, chest wall hernia, or thoracic instability, which may be common complications. If a second procedure must be performed, the patient must be aware of the complexity, risk of complications, and recovery period and have realistic expectations about the final results [12].

Alternatives for chest wall reconstruction

In all cases of chest wall reconstruction, stability, organ protection, and efficient mechanical ventilation must be the main goal. The skeleton and soft tissue may be replaced depending on the procedure; therefore, multiple materials and techniques

have been developed for a successful reconstructive outcome [13–15, 16].

Titanium-based systems have clear advantages over stainless steel- and ceramic-based systems, such as their biocompatibility, osseointegration, resistance to infection, and a high strength/weight ratio that supports body structures. Multiple titanium-based osteosynthesis systems are available with different reconstructive indications (Stratos/Stracos; MedXpert, Heitersheim, Germany); however, it has been reported that up to 44% of the implants fail at 1 year. They can be broken or displaced, which makes the incidence of failure very high and advocates for early removal whenever possible [14, 15, 16].

Soft-tissue reconstruction should not be discarded. A multidisciplinary team headed by a plastic surgeon must be assembled if needed for complex chest wall reconstruction. Soft-tissue reconstruction is based on the use of either free or pediculated myocutaneous flaps for external closure of the defect. The more frequently used muscles for chest wall reconstructions are the pectoralis major, latissimus dorsi, and rectus abdominis muscles [13, 15].

When a less-invasive reconstruction is mandated, a three-dimensional (3D) custom-made prosthesis can be implanted subcutaneously; it is a more-personalized approach to chest wall reconstruction proposed to fill up the defect resulting from the deformity of the bony structures with promising outcomes. Another alternative for defect correction is lipofilling using autologous fat [16].

The final stage of the reconstructive elevator in chest wall reconstruction is tissue bioprinting. Preliminary steps toward this goal are being taken in the form of new porous biomaterials that mimic bone's topological, mechanical, and mass transport properties. Promising results have been observed in the use of two-phase systems consisting of differentiation and growth factor-loaded nanoparticles embedded into printed biocompatible scaffolds, even for full-thickness chest wall defects.

Case report

A 21-year old man (88 kg, 1.92 m, body mass index = 23.8) with PE presented at the thoracic surgery department of the General Hospital of Mexico, "Dr. Eduardo Liceaga." With history of one previous attempt of surgical correction of PE at 18 years old, his clinical records described a video-assisted Nuss procedure and reported correction of thoracic defect by placing a retrosternal steel bar and a stabilizing metal bar to the 9th right side rib. Recovery in the intensive care unit (ICU) was required as part of the thoracic surgery protocol with an overall length of hospital stay of 4 days, after which he was discharged. The metal bar bent 2 weeks after surgery, requiring surgical removal. The patient was discharged from hospital 1 day after surgery and left untreated till 3 years later when he requested another consultation. His main complaint was esthetic, but other symptoms were described, such as exercise intolerance and mild chest pain.

A thoracic CT scan was performed, showing heart displacement and severe deformity of the thoracic wall contour. We calculated a Haller index of 3 points. Spirometry reported a restrictive respiratory pattern because of alteration of pulmonary parenchyma, thoracic wall compliance, and muscle insertions. The echocardiogram reported no ventricular function alteration, but there was a mild prolapse of the anterior portion of the mitral valve.

Surgery was performed under general anesthesia. A 10-cm parasternal incision was made with a scalpel, and tissue was dissected until reaching the anterior face of the concave-shaped sternum body. We disinserted the intercostal muscles until the costal cartilages of the sixth, seventh, eighth, and ninth ribs were exposed to present the deformity. Five millimeters of cartilage was excised bilaterally from all deformed cartilages to release the sternal body. Two skin incisions were performed at the lateral rib cage bilaterally at the site of previous scars to insert a titanium Lorenz bar (MedXpert). The transverse portion of the bar was allocated below the sternal

Fig. 1 **a** Anterior chest X-ray postsurgery in 2018. **b** Lateral chest X-ray postsurgery in 2018
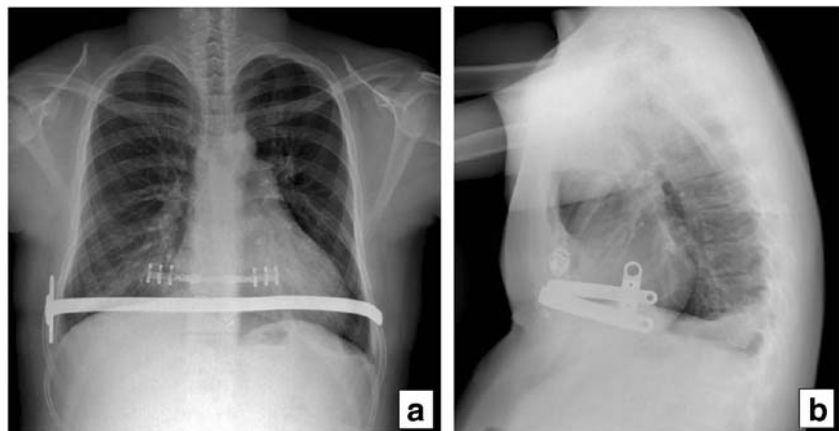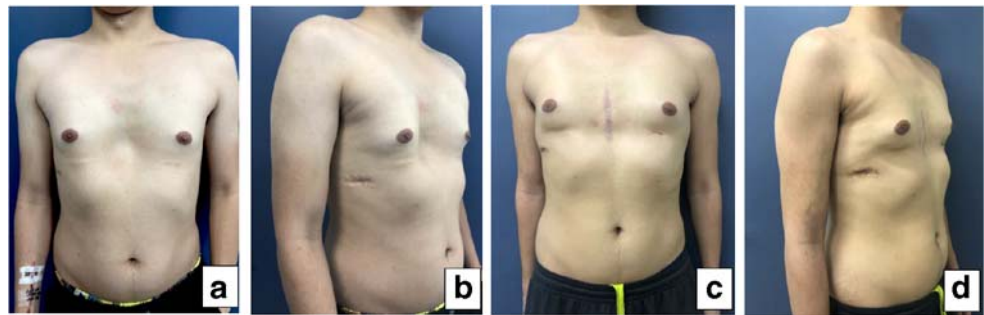
Fig. 2 a, b Presurgery photos. c, d One-year postsurgery photos

body to thereby correct the concavity. The bar was fixated to the right ninth rib with a metallic clip. The Lorenz bar was fixated with a retrosternal metallic wire so that it would not shift position. A second metallic bar (Stratos; MedXpert) was inserted below the sternum to unite and align ribs for extra stability (Fig. 1). A chest tube was placed through the left hemithorax and left at the site of the anterior mediastinum.

The patient showed adequate postsurgical evolution and required only 1 day in the ICU for vigilance. The chest tube was removed on the second day postsurgery. The patient was discharged from hospitalization on the fifth day with oral analgesics.

One-year postsurgery, the patient attended a follow-up consultation and clinical pictures were taken. They showed esthetic improvement regarding chest wall contouring (Fig. 2). A new thoracic CT scan was performed (Fig. 3), and the thoracic wall appears corrected, with the sternum, and both osteosynthesis materials fixated in place. The new Haller index was 1.9 points. The patient considered himself as asymptomatic and was pleased with the esthetic result. However, Lorenz bar removal is still pending, but it causes no discomfort.

Discussion

The surgical repair criteria for PE are well established depending on the clinical findings and cardiorespiratory symptoms developed by the patient. In older patients, however, their cosmetic disfigurement plays a more important role, which can itself become another indication of surgery. The appropriate timing of surgery is still being discussed. Usually, younger patients taken to a consultation by their parents are offered minimally invasive surgery with great overall results. However, older patients who sought consultation for themselves usually choose between surgical techniques depending on the available surgical expertise, cosmetic preference, and patient history. The patients' preferences should be considered once they have weighed the advantages and disadvantages of every procedure. The main reason for choosing an invasive procedure was not introducing a metal bar, while the second reason was the need for a second surgery using the Nuss procedure. However, the main reason for those who chose the Nuss procedure was the cosmetic result [4].

We are aware of newer, less-invasive techniques for chest wall reconstruction using more-flexible materials as biological meshes or 3D custom-printed silicone implants that can correct the chest wall contour with less morbidity [16]. In complex cases, however, such as severe PE with cardiac and pulmonary compromise and in previously treated and recidivated adult patients, skeletal reconstruction with more stable materials like titanium is a better choice.

We present the case of a young adult with a robust body type who was treated with MIRPE and recurred almost immediately. The patient's main concern was esthetic, but he was starting to develop severe symptoms from his deformity. We approached his case by combining both invasive and MIRPE techniques to resect the diseased and deformed cartilage,

Fig. 3 a Presurgery thorax CT scan. b One-year postsurgery thorax CT scan
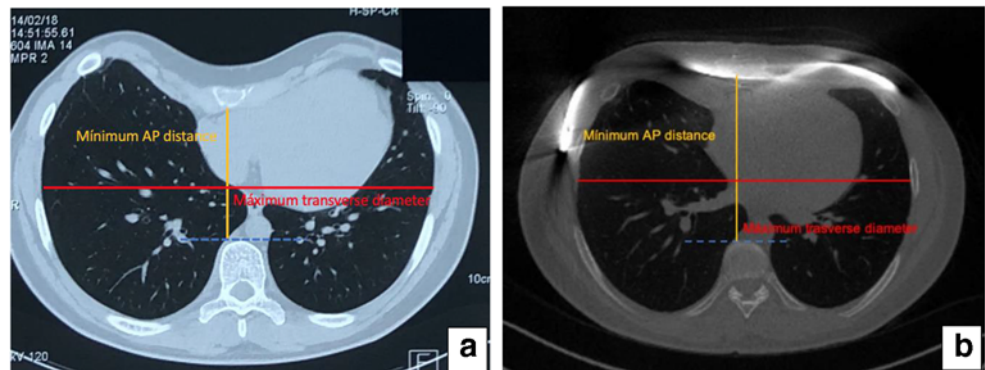
release sternum body, and project it forward using two different osteosynthesis systems to immediately and steadily correct his thoracic deformity.

Conclusion

The present case demonstrates the need for hybrid methods and more-aggressive approaches in complicated cases as shown by an adult patient with recidivating PE previously treated with MIRPE. A combined technique with resection of deformed cartilage and the placement of two steel bars as osteosynthesis for chest wall stabilization can achieve immediate correction of the defect to reduce cardiac and respiratory symptoms, but most importantly for the patient, to achieve a more-natural chest silhouette, which positively affects the patient's self-esteem and social life. The literature reports a few cases of the use of two steel bars for the correction of recurrent PE. The Stratos (MedXpert) system for thorax remodeling and stabilization, surgical experience, and long-term follow-up are still limited. In our case, we recognized that using osteosynthesis systems would not be enough to correct the deformity. Therefore, performing chondrectomy of deformed structures that contributed to the failure of the previous procedure and the stabilization of structures resulted in a better reconstruction outcome and prevented recurrence.

Compliance with Ethical Standards

Conflict of Interest   No

Funding   No

Ethics Committee approval   No, this is a case report and literature revision so no experiment was performed.

Informed Consent   We have informed consent for both surgery and the development of this paper.

Human and animal rights   This is a case report and literature revision so no experiment was performed , no animals were involved either.

References

1. Nuss D, Obermeyer R, Kelly RE Jr. Pectus excavatum from a pediatric surgeon's perspective. Ann Cardiothoracic Surg. 2016;5: 493–500.

2. Varela P. Pectus excavatum: history and new proposals for diagnosis and treatment. Rev Med Clin Condes. 2009;20:769–75.

3. Elsayed HH, Hassaballa AS, Abdel Hady SM, Elbastawisy SE, Ahmed TA. Choosing between the modified Ravitch and Nuss procedures for pectus excavatum: considering the patients perspective. Ann R Coll Surg Engl. 2016;98:581–5.

4. Nuss D, Kelly RE Jr, Croitoru DP, Katz ME. A 10-year review of a minimally invasive technique for the correction of pectus excavatum. J Pediatr Surg. 1998;33:545–52.

5. Fonkalsrud EW, Dunn JC, Atkinson JB. Repair of pectus excavatum deformities:30 years of experience with 375 patients. Ann Surg. 2000;231:443–8.

6. Kelly RE, Goretsky MJ, Obermeyer R, et al. Twenty-one years of experience with minimally invasive repair of pectus excavatum by the Nuss procedure in 1215 patients. Ann Surg. 2010;252:1072–81.

7. Croitoru DP, Kelly RE Jr, Goretsky MJ, Gustin T, Keever R, Nuss D. The minimally invasive Nuss technique for recurrent or failed pectus excavatum repair in 50 patients. J Pediatr Surg. 2005;40: 181–6.

8. Redlinger RE Jr, Kelly RE Jr, Nuss D, Kuhn MA, Obermeyer RJ, Goretsky MJ. One hundred patients with recurrent pectus excavatum repaired via the minimally invasive Nuss technique-effective in most regardless of initial operative approach. J Pediatr Surg. 2011;46:1177–81.

9. Pilegaard HK. Extending the use of Nuss procedure in patients older than 30 years. Eur J Cardiothorac Surg. 2011;40:334–7.

10. Luu TD, Kogon BE, Force SD, Mansour KA, Miller DL. Surgery for recurrent pectus deformities. Ann Thorac Surg. 2009;88:1627–31.

11. Johnson KN, Jaroszewski DE, Ewais M, Lackey JJ, McMahon L, Notrica DM. Hybrid technique for repair of recurrent pectus excavatum after failed open repair. Ann Thorac Surg. 2015;99: 1936–43.

12. Jaroszewski DE, Ewais MM, Lackey JJ, et al. Revision of failed, recurrent or complicated pectus excavatum after Nuss. Ravitch or cardiac surgery. J Vis Surg. 2016;2:74.

13. Novoa NM, Aranda Alcaide JL, Gomez Hernández MT, Fuentes MG, Goñi E, Jimenez Lopez MF. Chest wall- reconstruction: yesterday, today and the future. Shanghai Chest. 2019;3:15.

14. Corcoles Padilla JM, Bolufer Nadal S, Kurowski K, Gálvez Muñoz C, Rodriguez Paniagua JM. Use and versatility of titanium for the reconstruction of the thoracic wall. Cir Esp. 2014;92:89–94.

15. Dagnino Urrutia BL, Patillo Silva JC, Salisbury Devizenci C, Cifuentes Ortiz IJ. Thoracic wall reconstruction with latissimus dorsi flap in pediatric patients.Cir Plast Ibero-Lat.2019;45:67-72.

16. De Baerdemaeker RA, Hendrickx B, Geeroms M, Zeltzer AA, Hamdi M. Correction of Pectus Excavatum anterior chest wall deformity in adults using custom made silicone implant. Ann Plast Reconstr Surg. 2019;2:1021.