 Cir Pediatr. 2021; 34: 171-179

Chest wall malformations: a new perspective in diagnosis and treatment

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INTRODUCTION

In 1998, the publication of the original article by Nuss et al. changed pectus excavatum (PEX) treatment forever[1], since this revolutionary minimally invasive technique offers excellent short- and long-term results. As a result of this, the technique became popular and currently stands as the best alternative to repair this deformity. Patients were so excited with the results they decided to undergo PEX surgical repair according to Nuss procedure.

As a result of the increase in cases, the number of scientific papers on the subject also soared. Most of them focused both on physiopathology and technical improvements to prevent and/or reduce complications inherent to surgery[2-4].

This update provides an overview of the most important advances made in PEX physiopathology regarding the role of cardiac compression. It also describes those changes that have helped prevent and/or reduce complications such as cardiac perforation, bar migration, insufficient repair, and postoperative pain.

CARDIAC COMPRESSION AS A KEY FACTOR

PEX was considered as a merely esthetic issue for years –and still is today[5]. This is due to the fact evidence of its functional impact is inconclusive[6,7]. Intolerance to stress and dyspnea, which are observed in a great amount of PEX patients, cannot be solely explained by pulmonary function test findings[8-10]. On the other hand, even though cardiac compression can be clearly noted during thoracoscopic PEX repair and could well explain clinical symptoms, PEX is rarely identified by echocardiographic studies. In the words of Claude Bernard, “The experimenter who does not know what he is looking for does not understand what he finds.” And conventional echocardiography professionals have been trained to assess other types of cardiac impact.

Various studies evaluating cardiovascular impact using various methodologies such as echocardiography with and without stress, transesophageal echocardiography, cardiac MRI (CMRI), and cardiopulmonary tests have been carried out.

Over time, it has been increasingly demonstrated that PEX compresses the heart, especially the right cavities, which causes functional repercussion. In a study carried out in 99 patients undergoing MRI and cardiopulmonary stress test, PEX severity was associated with decreased cardiac output[11].

Additionally, some studies have demonstrated cardiac compression reversion following PEX repair using imaging tests such as intraoperative transesophageal echocardiography[12-14]. Increased RV ejection fraction one year following surgery has also been demonstrated[15].

In terms of cardiopulmonary tests, results have improved following PEX repair both in children and in adults[9,16].

CMRI has been used in studies examining cardiac dysfunction in PEX. This is partly due to the fact the suboptimal ultrasound window limitations involving echocardiography in a relatively large number of PEX patients do not occur[17-20]. CMRI has allowed cardiac compression at rest to be classified into three groups: type 0 (no compression), type 1 (RV compression), and type 2 (RV and auriculoventricular groove compression) (Fig. 1). In this study, which included 60 patients, 77% had RV compression, with compression type 1 being more frequent (45%)[21]. This classification then allowed CMRI, echocardiography under stress, and chest CT-scan to be assessed in 59 patients, which led to two conclusions: 1) the greater compression

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Date of submission: Julio 2020 *Date of acceptance:* Julio 2020

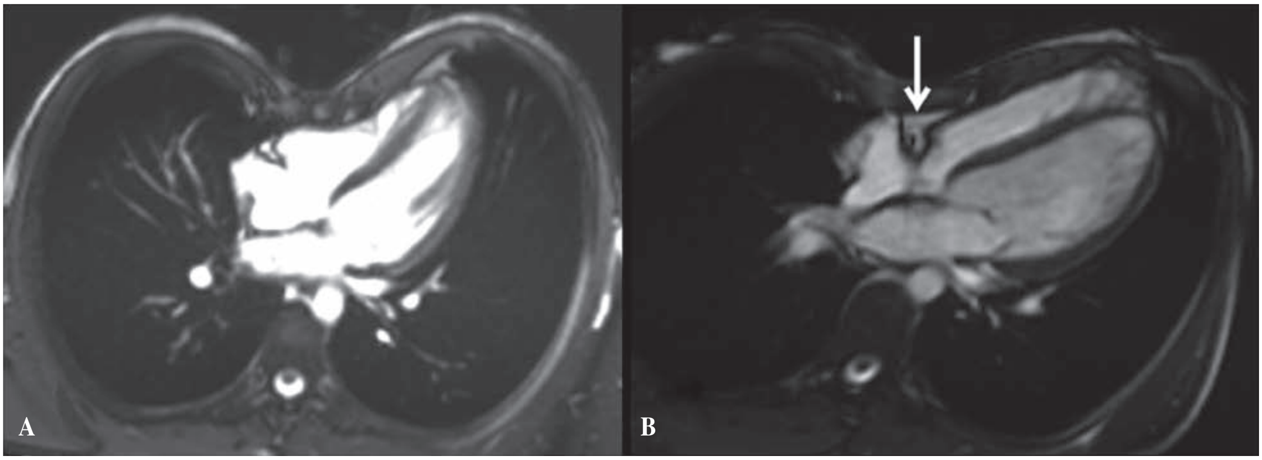

Figura 1. RMC de dos casos de PEX con compresión cardiaca. En A puede observarse que la compresión se limita a la pared libre del ventrículo derecho (tipo 1); en cambio, en B, el surco auriculoventricular (flecha) también se encuentra comprometido (tipo 2).

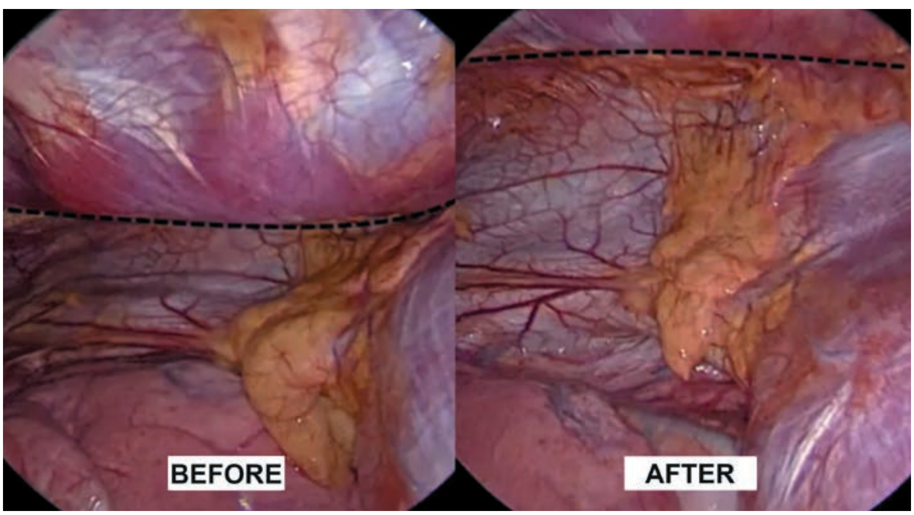

Figure 2. Thoracoscopic view of the relationship between the sternum and the heart during Nuss procedure. On the left, the dotted line follows the internal side of the sternum compressing the heart prior to sternal elevation; and on the right, following sternal elevation, the line is separated, creating a real safety space for precardiac dissection.

at CMRI was, the more significant right ventricle diastolic dysfunction proved to be; and 2) compression levels were associated with PEX severity indexes[22]. In a series consisting of 269 PEX patients undergoing echocardiography under stress –the largest up until now–, 64% had abnormal septal movement, 29% had RV diastolic dysfunction, and 16% had RV systolic dysfunction[23].

In conclusion, even though the role of the respiratory and muscular systems should not be underestimated, that of cardiac compression has been clearly supported by evidence and has become a new surgical objective in PEX repair.

MINIMALLY INVASIVE PEX REPAIR IS NO LONGER A DANGEROUS SURGERY

In light of the complications associated with Nuss procedure[2-4,24-28], some healthcare facilities have implemented changes to the original technique[29-31], such as thoracoscopic visualization, retrosternal subxiphoid dissection, and sternal elevation.

Sternal elevation stands as the most important change for the authors, since it allows a dissection plane to be created between the posterior side of the sternum and the heart, thus reducing the possibility of cardiac perforation (Fig. 2). In addition, sternal elevation allows for a "temporary repair of the defect", which prevents the defect from being repaired when passing the "pectus introducer" – nowadays known as "saber" or "samurai" (Figs. 3). Before sternal elevation was implemented, intercostal muscle strain used to be frequent as a result of having to apply significant force when passing from side to side. Thanks to sternal elevation, this complication has entirely disappeared in our practice.

Sternal elevation varies according to the various surgical groups. Schier[32] described the use of a vacuum bell,

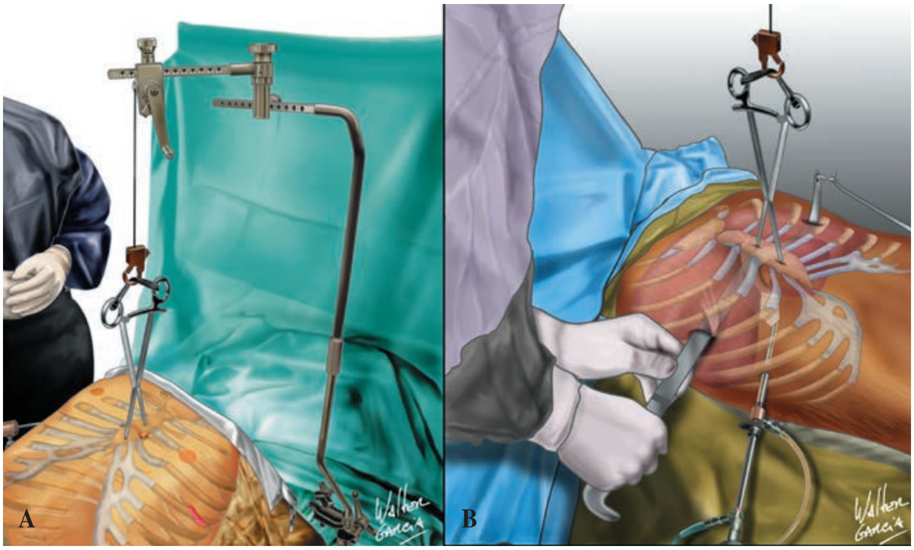

Figura 3. A) Se esquematiza la elevación esternal con un una grúa Rultract y una pinza de Lewin colocada en sentido longitudinal al esternón en un paciente con *pectus excavatum* durante la reparación. B) Una vez elevado el esternón, se puede observar el pasaje de un "samurái" o *pectus introducer* para realizar la disección del espacio retroesternal y facilitar el pasaje del implante.

Jaroszewski combined a Rultract crane with a Lewin clamp (Lewin Spinal Perforating Forceps)[33,34], and Park reported the use of a crane integrated with the operating table and connected with a wire suture fixated to the sternum[35]. Recently, Park proposed a screw-based sternal elevation technique instead of the wire suture technique[36].

Bar migration, either as a result of rotation, lateral displacement, or depression[2,4], is another major complication. Even though migration may require re-intervention, rotation implies a risk of lesion for vital structures such as the large vessels, with fatal consequences[26]. To prevent the bars from migrating, lateral stabilizers were added to the original technique[29]. Many groups use unilateral or bilateral stabilizers fixated to the ribs using a wire[29]. However, various publications still report bar migration and wire rupture[37-40].

With the purpose of minimizing rotation and migration risks, Park[41] described the use of lateral bridges in 80 patients with a mean age of 17.5 years (range: 6-38 years), and with an efficacy of 100%. Following Park concepts, we recently published our experience with the use of a self-blocking bridge system since 2016[42]. As an additional advantage, the use of bridges prevents the bar from being fixated to the ribs and the muscle wall, and it entirely avoids axial and lateral bar rotation. In the case of crossed bars, this system is made up of an entire variety of curved bridges adapted to all bar configurations, according to each case.

FULL CHEST REPAIR *VS.* REPAIR OF THE MOST DEPRESSED SITE

Both the original description of minimally invasive PEX repair (MIPER) and Haller index[43] have focused on the most depressed site of the rib cage in order to establish diagnosis and severity. However, as it can be observed in Kelly et al.'s classification[44], PEX clinical presentation is very heterogeneous (Figs. 4), which has led to other indexes, such as the "repair index" – which considers the distance between the anterior rib line and the most depressed site of the rib wall[45] –and the "Titanic index"[42]– which takes into account the percentage of the sternum located behind the anterior rib line.

In addition, the chest can be either symmetrical or asymmetrical, which determines bar shape and bar entrance and exit sites in the chest[46] (Fig. 5).

When assessing surface morphology using the optical scanner pre- and post- MIPER, a complete remodeling of the rib cage is noted, with increased transversal and anteroposterior chest diameters – at the expense of rib horizontalization. In addition, other favorable changes in patient position such as reduced shoulder flexion, dorsal kyphosis, and head anteriorization (Fig. 6) may be observed.

The sandwich technique –which allows highly rigid asymmetric deformities to be repaired using a set of intra- and extra-thoracic bars–, the magic string –which is used to flatten anterior chest wall protrusions–, and the flare buster –which shapes the associated rib flares[47]– can complement MIPER with excellent results.

To sum up, the objective of PEX repair should be full chest repair and not simply the repair of the most depressed site of the chest. This has a direct influence on the shape, length, location, and amount of bars to placed, and requires a customized approach in each case.

We recently published the customized perioperative process of 130 surgical patients with PEX, which includes preoperative planning based on CT-scan 3D chest reconstruction, preoperative test of semi-automatically designed

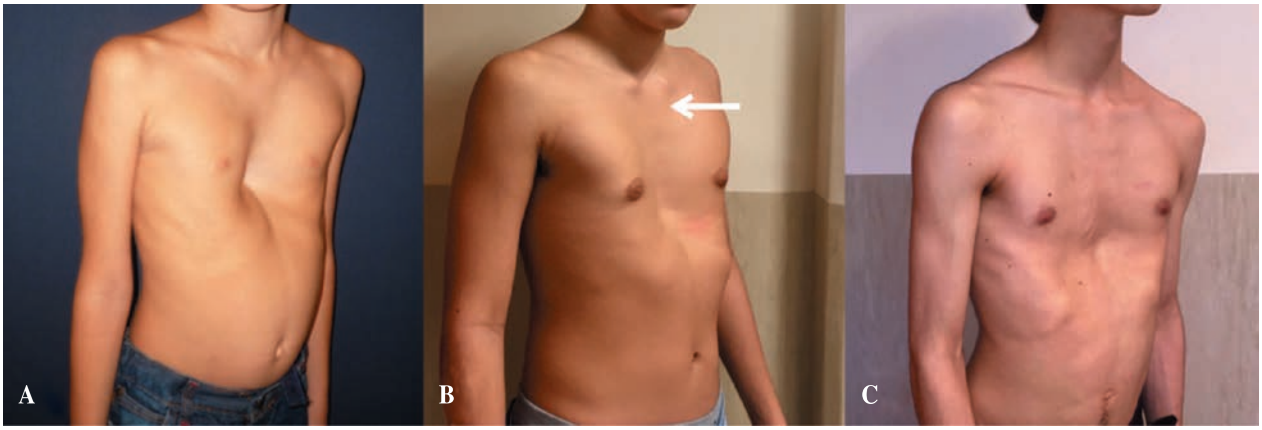

Figura 4. Se observan tres pacientes con deformidad excavada del tórax de distinto tipo. A) Deformidad tipo "punch" severo. B) Deformidad tipo "gran cañón". Nótese que el hundimiento comienza en el tercio superior del esternón (flecha). C) Deformidad tipo "plato", en el que toda la cara anterior de la pared costal se encuentra aplanada. Estos pacientes suelen presentar asociación de restricción ventilatoria y compresión cardiaca.

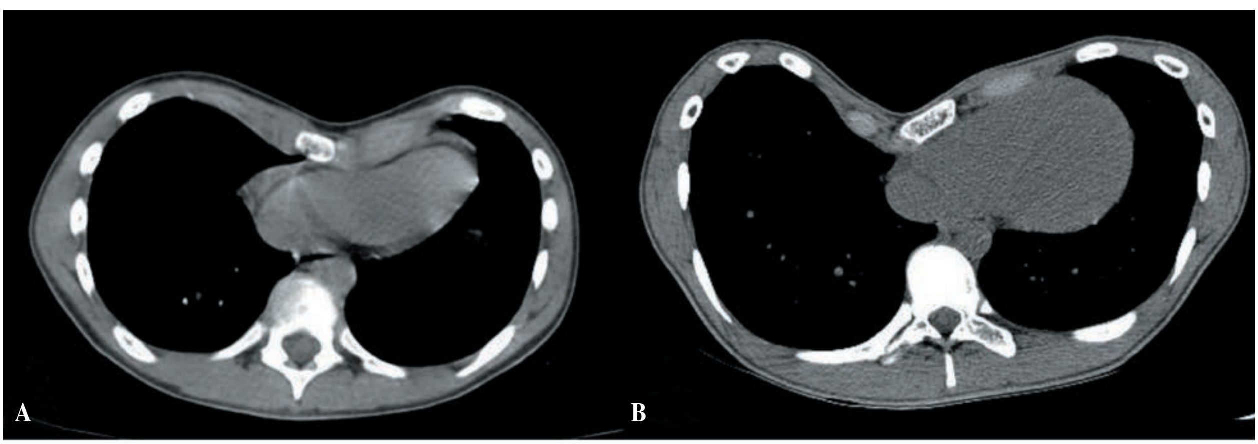

Figura 5. La simetría de la pared costal debe ser considerada al diseñar los implantes retroesternales en un caso de PEX. Nótese la marcada diferencia entre (A) un paciente simétrico, y (B) un paciente asimétrico con PEX.

customized templates, and bar customization[42]. Many repairs have been carried out in patients with significantly different clinical presentations using this procedure, which has demonstrated to be consistent over time (Fig. 7). These concepts have also been applied in patients with complex chest wall malformations such as pectus arcuatum or Poland syndrome[48] (Fig. 8).

CRYOANALGESIA FOR POSTOPERATIVE PAIN MANAGEMENT IS NO LONGER A PAINFUL SURGERY

An undesired effect of MIPER is postoperative pain, which requires the administration of high doses of opioids and increases hospital stay in various days. Several management strategies including epidural anesthesia, intercostal blockade, and patient-controlled analgesia[49-54] have been proposed.

The first reports on the use of cryoanalgesia for postoperative pain control in PEX demonstrated the need for a lower amount of opioids and a significant reduction in hospital stay, catching the attention of groups specialized in chest deformity surgery[55-59]. In 2016, Keller et al. published a comparative, retrospective study of 26 patients undergoing intercostal cryoanalgesia during MIPER, and 26 patients receiving epidural anesthesia, which revealed a significant reduction in hospital stay (3.5 ± 0.83 days *vs.* 5.79 ± 0.93 days, $p < 0.001$) and need for intravenous narcotics (49.0 mg ± 32.7 *vs.* 119.8 mg ± 95.1, $p = 0.0011$) in the cryoanalgesia group[56]. In 2019, this finding was confirmed in a randomized study

Figura 6. En el A, un paciente con PEX parado de perfil en el que se observa la típica cifosis dorsal, antepulsión de hombros y cabeza adelantada. En B, el mismo paciente, dos años después de una toracoplastia con tres implantes retroesternales. Nótese la corrección de la postura con resolución de la cifosis y alineamiento de cabeza y hombros.

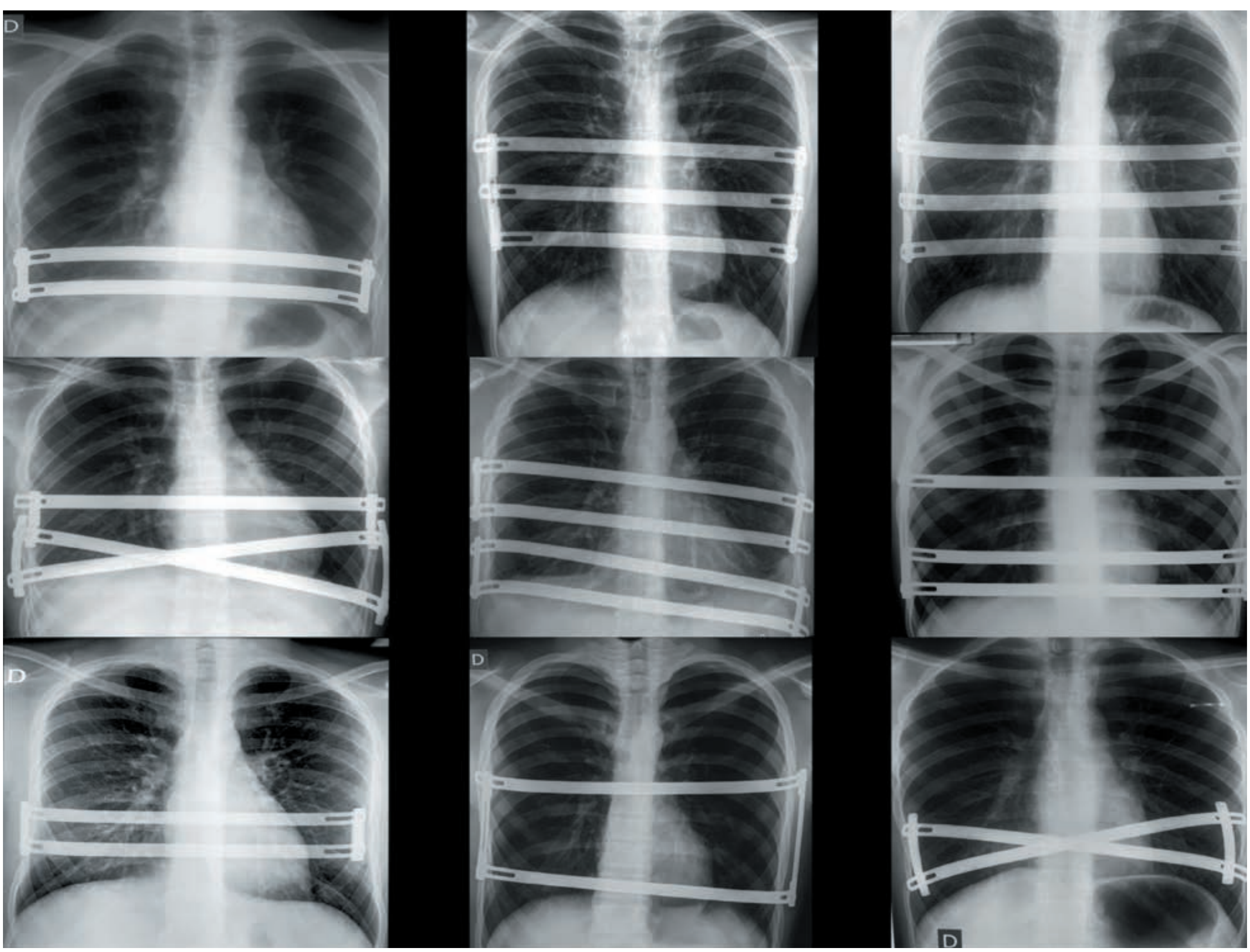

Figure 7. A wide array of possibilities in the number and location of bars and lateral bridges is observed. The heterogeneity of chest wall deformities requires different types of prostheses, and preoperative planning is essential for an optimal result. Re-printed from the Journal of Pediatric Surgery, vol. 55(12), Bellia-Munzón G, Martínez J, Toselli L, et al. From bench to bedside: 3D reconstruction and printing as a valuable tool for the chest wall surgeon, pages: 2703-2709, 2020, authorized by Elsevier.

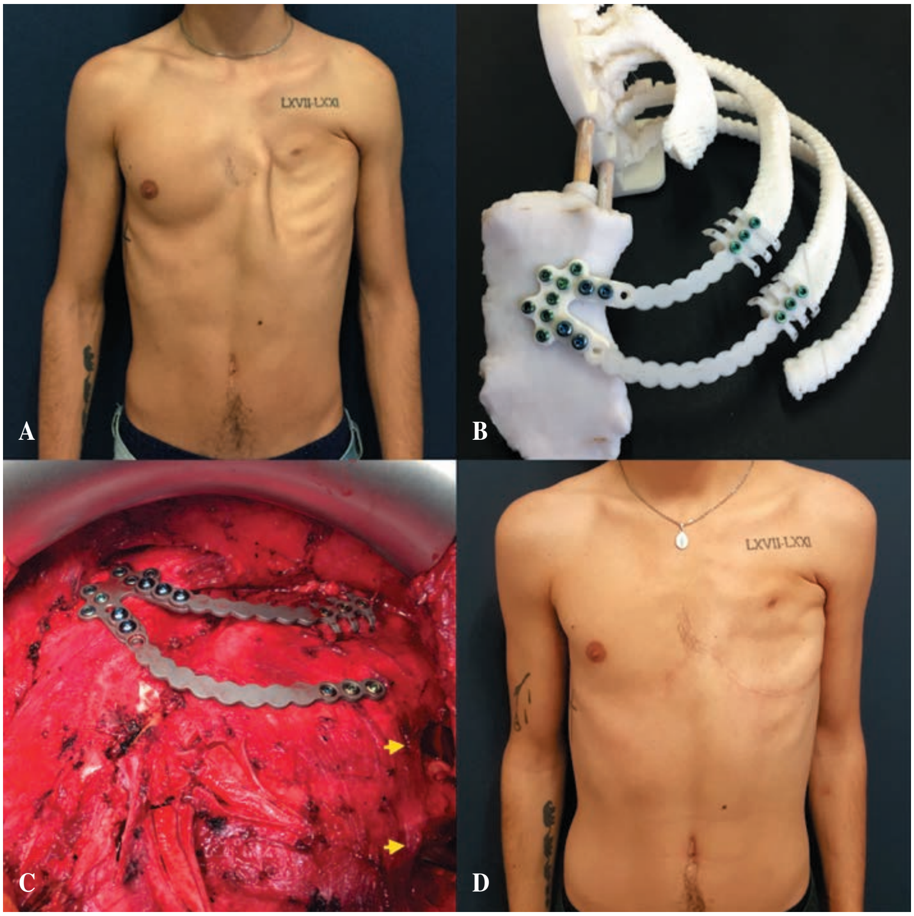

Figura 8. Proceso de simulación usando tecnología tridimensional y reparación híbrida con prótesis customizadas de un síndrome de Poland complejo. A) Se evidencia el aspecto preoperatorio del pecho del paciente con síndrome de Poland con agenesia parcial del esternón, agenesia condral e hipoplasia costal. B) Simulación de la reparación utilizando un modelo impreso en 3D con una escala 1:1. C) Una fotografía intraoperatoria demuestra el campo quirúrgico con costillas de titanio con agarre costal. La costilla inferior no posee agarre costal porque fue considerado innecesario durante la simulación. Dos cabezas de flecha marcan dos implantes retroesternales emergiendo del tórax en el lado izquierdo. D) Se observa el aspecto del paciente dos años después de la cirugía. Reimpreso de The Annals of Thoracic Surgery, journal pre-proof, Martínez J, Toselli L, Giménez Aleixandre Cristina, y cols. Surgical Planning, Simulation, and prostheses customization for complex chest wall malformations, con permiso de Elsevier.

of two 10-patient groups undergoing MIPER –one with cryoanalgesia and the other with epidural anesthesia-, which showed a median reduction in hospital stay from 5 to 3 days using cryoanalgesia (Mann-Whitney U, $p = 0.0001$) and a 52-82% reduction in the amount of opioids required in the first three postoperative days ($p < 0.01$ each day)[55].

Cryoanalgesia involves the application of cold for axonotmesis purposes, i.e., the destruction of the axon and the myelin sheath of peripheral nerves with preservation of the connective sheaths. Nerve destruction occurs in the trajectory exposed to cold application, generally between -60ºC and -80°C (Fig. 9). Nerve regeneration occurs from the nerve sides divided on to the connective sheaths, with full restoration being achieved in approximately six weeks – while freezing 2 cm in length.

Although this is not always the case, preoperative and postoperative pregabalin or gabapentin are frequently indicated to prevent neuropathic pain. However, a recent study assessing the incidence of neuropathic pain in 30 patients under 21 and in 13 patients over 21 both undergoing cryoanalgesia during MIPER without preventive medication found neuropathic pain in 3 adults only, with the authors suggesting further studies are required to establish whether this adverse effect is present in young patients or not[60].

The approach can be intrathoracic at surgery itself or percutaneous in a simultaneous or previous procedure. Neither strategy has proved superior to the other.

Recently, a cohort of 23 patients undergoing cryoanalgesia during MIPER from September 2018 to April 2019 was published, with a mean hospital stay of 1.64 ± 0.73 days[61].

Cryoanalgesia is probably the best postoperative pain management strategy. Given its high success rates, we believe it will be increasingly used.

CONCLUSIONS

This update provides an overview of the most important advances made in PEX, including the role of cardiac compression in PEX physiopathology, the decrease in complications as a result of sternal elevation, the need for full chest repair as part of the treatment, and the use of cryoanalgesia for postoperative pain management.

Figure 9. During intrathoracic cryoanalgesia, selecting the adequate entry site for the cryoanalgesia probe is key to adequately reach all target intercostal spaces. The intrathoracic thoracoscopic view shows that the angulated tip of the probe is leaning parallel to the lower side of the upper rib.

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