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Life-threatening cardiovascular adverse events related to pectus excavatum surgery

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Abstract

Background: The occurrence of life-threatening cardiovascular adverse events related to pectus repair is underestimated.

Methods: Literature review of severe adverse events affecting the heart, pericardium and large vessels of open or minimally invasive repair.

Results: In patients undergoing Willital-Hegemann repair, we identified one death caused by intraoperative cardiac arrest. In patients undergoing Ravitch-type repair, we identified 11 non-lethal life-threatening cardiovascular adverse events: two per/postoperative cardiac injuries and six delayed intracardiac bar migrations (overall, six successful cardiac procedures under CPB were performed); and three intrapericardial migrations of broken sternal wire and consecutive tamponade treated by wire removal and pericardial drainage. Finally, we identified one aortic injury caused by a broken sternal wire 28 years after sternal turnover.

In patients undergoing minimally invasive repair we identified 27 cardiac injuries during procedure and three later Nuss-bar removal; one intracardiac migration of the Nuss-bar (overall, four patients died and 11 underwent cardiac procedures under CPB); three procedural vena cava injuries (one patient died); six obstructions of the inferior vena cava (n=3) or right ventricular outflow tract (n=3); three episodes of major bleeding from the ascending aorta (one patient died); two pericardiectomies and three late-onset cardiac tamponades. Eight patients sustained procedural or late-onset cardiac arrest (five patients died). Overall, we identified 56 life-threatening cardiovascular adverse events requiring 12 cardiac/aortic procedures under CPB. There were 11 deaths and two cases of severe hypoxic brain injury.

Conclusions: During pectus surgery, the cardiovascular morbidity/mortality related to minimally invasive repair appears more severe, in comparison to open repair.

Introduction

Pectus Excavatum (PEx) is the most common anterior chest wall deformity. Its pathogenesis is still controversial. However, overgrowth and/or distortion of costal cartilage play a key role in the pathophysiologic and clinical features of the disease [1]. The deformity usually worsens as the child grows [2]. While the indication for surgical management is mainly cosmetic it also aims at improving patients' quality of life [3] and exercise performance [4].

Management of complex chest wall deformities such as PEx has improved significantly over the past 50 years with high long-term success rates. However, following these procedures some life-threatening adverse events have been reported, mainly cardiovascular injuries. Due to the chest deformation, the anatomical ratios are modified, which facilitates the occurrence of heart or great vessel injury. Furthermore, other lesions can occur in the mid/long term due to steel wire or metal bar fracture and/or dislocation. Such cardiovascular adverse events are even less acceptable since this surgery is performed for cosmetic or psychological purpose in the majority of cases.

There are two types of PEx surgery: open and minimally invasive repair. The most usual open repairs are the Ravitch-type procedures based on the subperichondrial resection of abnormal cartilages combined with a transverse osteotomy across the anterior table of the upper sternum and the routine use of a metallic support bar to maintain the elevated sternum. This support bar is usually removed at six months [5-8]. One variant is the Willital-Hegeman technique, in which three metallic bars are used to stabilize the remodeled anterior chest wall [9]. Finally, the "sternal turnover", a 180° reversal on its axis of the chondrosternal plastron after upper sternal section [10] is an operation currently abandoned.

The minimally invasive repair (or close repair) is commonly referred to MIRPE (Minimally Invasive Repair of Pectus Excavatum), or Nuss procedure [11]. Depending on the deformity, it consists of placing one or more preformed curved bars in the thoracic cavity under thoracoscopic control. Then a 180° turning manoeuver of bar corrects the concavity of the anterior chest wall.

Based on literature data, the purpose of this study is to assess the occurrence of Life-Threatening Cardiovascular Adverse Event (LCAE) during PEx surgery, in both open and minimally invasive approaches.

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Material and methods

This review was designed according to the recommendations for systematic reviews of the literature and meta-analyses [12]. Bibliographic searches were conducted using the Pubmed, Medline, CINAHL, Cochrane and Embase bibliographic and bibliometric databases, including the following keywords: Pectus excavatum, close repair, Nuss, MIRPE, open repair, Ravitch, sternochondroplasty, complications, cardiovascular complications; including studies of prospective and retrospective series, case reports and case series, with cross-referencing of bibliographical references.

We selected the articles reporting or mentioning severe adverse events affecting the heart and/or large vessels, during PEx repair. Procedural pericardial contusion or laceration, or early postoperative effusion were not taken into account. On the other hand, we selected as LCAEs the delayed pericardial events such as constriction or late-onset tamponade. Cardiac rhythm disorders and damage to peripheral vessels (mainly the internal thoracic artery) were not taken into account.

A total of 531 medical articles were examined and 67 were selected, reporting cardiovascular adverse events severe enough to trigger life-threatening or fatal prognosis. Articles published in English, French and Spanish were included. Additionally, we selected two articles from the non-medical literature, and took into account duplicate and triplicate studies reporting the same case of LCAE.

Results

Pectus excavatum open repair

After PEx open repair, we identified 13 LCAEs including one lethal case, as summarized in Table 1. This death was reported by Saxena and coworkers [9] in their series of 1262 patients treated by the Willital-Hegemann procedure as a result of a peroperative cardiac arrest in a patient with an unspecified history of "cardiac abnormality".

Procedural and postoperative lesions (n=2)

The unique procedural cardiac injury was reported by Hernandez and coworkers [13]. In a child with a past history of neonatal cardiac surgery the injury occurred during an attempt of Ravitch-type repair in infancy. The definitive cure of PEx was performed at the age of 16 by means of Nuss-bar placement through sternotomy and Cardiopulmonary By-Pass (CPB) for safety reasons.

In the postoperative period, we identified an iatrogenic cardiac injury in a seven-year-old child who had undergone a PEx open repair using Kirchner wires to stabilize the remodeled anterior chest wall. Following blind replacement of a dislocated wire protruding beneath the skin on Day 5, he sustained severe congestive heart failure. Emergent surgical exploration showed a pierced right atrium, a torn septal leaflet of the tricuspid valve and noncoronary aortic cusp, and a large traumatic ventricular septal defect. Lesions were repaired under CPB. However, the child showed residual tricuspid and aortic regurgitation as sequelae [14].

Intracardiac migration of metal support bar (n=6)

We identified two cases of early displacement (within six months) of a metal support bar in two young adults. One bar penetrated the right and left ventricle across the interventricular septum [15] and the other induced an aorta-to right ventricular fistula [16]. In both cases, the complication was caused by the use of a too short bar, thus facilitating unintended migration into the mediastinum. Four other

Table 1. Open repair. Characteristics and outcomes for 13 patients who sustained LCAE

Authors (Procedure)	Patient age (y) Past history	Cardiac/Aortic injury (Delay)	Approach for injury repair (CPB)	Outcomes
Saxena [9] (Wilital-Hegemann)	(NM)	Procedural unintended cardiac arrest in a patient with "unspecified cardiac abnormality" (no cardiac injury)	None	Dead
Hernandez [13] (Ravitch-type)	(NM). Neonatal cardiac surgery	Procedural non-lethal cardiac injury during attempt of Ravitch-type repair in infancy	NM	Nuss-bar under CPB at 16
Pircova [14] (Open repair + Kirchner wires)	(7)	Right atrium, tricuspid valve, traumatic ventricular septal defect (blind replacement of a Kirchner wire) (Day 5).	Sternotomy (CPB)	Tricuspid and aortic regurgitation
Intracardiac migration of metallic support bar				
Elami [17] (Ravitch-type)	(12)	Migration of broken metal bar into the right atrium (2 years)	Sternotomy (Steady state CPB)	Recovery
Dalrymple-Hay [15] (Ravitch-type)	(19)	Bi-ventricular perforation due to metal bar migration (6 months)	Sternotomy (CPB)	Recovery
Onursal [18] (Ravitch-type)	(18)	Migration of broken metal bar into the right ventricle (4 years)	Clamshell incision (Steady state CPB)	Recovery
Aydemir [16] (Ravitch-type)	(23)	Rupture of the aortic sinus of Valsalva + Aorta-to-right ventricular fistula, due to metal bar migration (1 month)	Sternotomy (CPB)	Recovery
Zhang [19] (Ravitch-type)	(53)	Migration of 2 metal bars into the right ventricle, and pulmonary trunk+ left main bronchus (37 years)	Left anterior thoracotomy (CPB)	Recovery
Biliboni [20] (Ravitch-type)	(25)	Metal bar migration into the right ventricle and through the tricuspid valve (8 years)	Sternotomy (CPB)	Mild tricuspid regurgitation
Migration of broken sternal wire				
Barakat [21] (Ravitch-type)	(24)	Pericardial tamponade due to broken sternal wire with pericardial/ epicardial injury (2 years)	Sternotomy	Recovery
Cope [22] (Ravitch-type)	(24)	Pericardial tamponade due to broken sternal wire with pericardial injury (2 years)	Thoracotomy	Recovery
Lee [23] (Hybrid procedure)	(15)	Pericardial tamponade due to broken sternal wire with pericardial injury (1 year)	NM	Recovery
Mieno [24] (Sternal turnover)	(34)	Migration of broken sternal wire into the ascending aorta (28 years)	Sternotomy, Prosthetic replacement of the ascending aorta (CPB)	Recovery

LCAE: Life-Threatening Cardiovascular Adverse Event; NM: Not Mentioned; CPB: Cardiopulmonary ByPass

patients sustained intracardiac bar migration from two to 37 years after the initial procedure as a consequence of failure to remove the bar in a timely manner and/or broken material [17-20]. Injured cardiac structures are detailed in Table 1. Among these six patients, four underwent cardiac surgery under CPB and steady-state CPB in two cases. All these patients showed satisfactory results.

Migration of broken sternal wire (n=4)

In three patients, intrapericardial migration of a broken sternal steel wire (n=2), or steel wire used for fixation of the retrosternal metallic bar in the midline during a hybrid technique (Ravitch + Nuss-bar), induced a hemopericardium and consecutive tamponade. Healing was achieved by draining blood, and removing the broken wire [21-23].

Finally, 28 years after sternal turnover the aortic migration of a broken sternal wire generated lesions severe enough to require prosthetic replacement of the ascending aorta under CPB [24].

Minimally invasive repair of pectus excavatum

After MIRPE, we identified a total of 56 LCAEs that resulted in 11 deaths, and two cases of severe hypoxic brain injury as a consequence of procedural cardiac arrest.

Procedural heart injury (n=27)

A heart injury may occur when inserting the introducer or the Nuss-bar into the thoracic cavity, or when repositioning a dislocated bar. We identified 27 intraoperative cardiac perforations or lacerations [11,13,25-30,32-47]. Nine patients had a history of cardiac surgery. Three were treated for PEx recurrence, two after MIRPE [28,43]. and one after Ravitch-type repair [34], respectively.

The cardiac injury resulted in death of three young patients [19,29,37], and severe hypoxic brain injury in one [34]. Characteristics of patients are summarized in Table 2. Some data are lacking in five cases. Among the 22 other patients, PEx treatment was definitely discontinued in five. Finally, one patient underwent a later Nuss-bar placement under CPB for safety reasons [14].

Trans myocardial migration of the Nuss-bar (n=1)

One year after MIRPE, a patient sustained a trans myocardial migration of the Nuss-bar, discovered by an intense cardiac murmur audible by the patient himself. Removal of the bar and repair of considerable cardiac damages required a 14-hour CPB procedure, with a good result at two years, except for the absence of PEx correction. This case was reported in a non-medical journal [48].

Cardiac injury during later Nuss-bar removal (n=3)

Bouchard and coworkers [34] reported a lethal heart injury during Nuss-bar removal due to fibrotic adhesions between the bar and the pericardium, as a consequence of postoperative pericardial effusion. The same mechanism in a patient with a history of cardiac surgery for transposition of large vessels and mild pulmonary valve stenosis (Mustard technique) resulted in a double ventricular injury and massive bleeding [49]. Finally, a tear of the right ventricle with massive hemorrhage was reported by Sakakibara and coworkers [50]. The ventricular lesion was repaired under CPB. Retrospective analysis of the sagittal sections of the CT scan performed prior to the Nuss-bar removal showed evidence of intra ventricular bar migration.

Vena cava injury or occlusion (n=6)

Three patients suffered a procedural injury of the superior (n=1) or inferior vena cava (n=2). No information was provided about the case of superior vena cava injury [51]. An inferior vena cava injury occurred during a redo-MIRPE, which was repaired by sternotomy [43]. Last, another case of procedural inferior vena cava transfixion occurred, leading to cardiac arrest and patients' death after a long coma [52].

Three cases of acute obstruction of the inferior vena cava were reported [31,53,54]. At the end of the procedure, patients exhibited hemodynamic instability, oliguria and abdominal distension. An exploratory laparotomy was performed in two patients [31,53]. The diagnosis of acute obstruction of the inferior vena cava (probably as the consequence of a plication secondary to a sudden change in chest wall and diaphragm geometry) was made by cavography, ultrasound or CT scan. Although one patient sustained a procedural cardiac arrest [54], removal of the Nuss-bar was life-saving. However, no subsequent PEx correction was performed in any patient.

Ascending aortic lesions (n=3)

Two months after MIRPE, the dislocation of the Nuss-bar in the cranial direction induced an hemopericardium due to a tear of the ascending aorta. After emergent sternotomy, the pericardium was emptied of blood. After careful removal of the Nuss-bar, hemostasis of the ascending aorta was achieved with pledgetted sutures [55]. A similar cranial dislocation caused a false aneurysm of the ascending aorta, which ruptured when the Nuss-bar was removed. A prosthetic replacement of the ascending aorta under CBP and deep hypothermia circulatory arrest was successfully performed [56].

Finally, a forensic medical case of lethal aortic hemorrhage during Nuss-bar removal was reported in a legal publication [57].

Stenosis of the pulmonary artery or right ventricular outflow tract (n=3)

Two cases of stenosis of the pulmonary artery trunk [58] or right ventricular outflow tract [59], compressed by the curved left sided extremity of the Nuss- bar occurred as a result of a right lateral shift of the Nuss-bar. Symptomatology with dyspnea and/or systolic murmur retroceded after Nuss-bar removal, but the former patient showed tricuspid valve insufficiency as sequel [58]. Finally, 11 years after MIRPE, a 23-year-old patient who experienced a syncopal episode was found to have a retrosternal fibrous callus that severely compressed the right ventricle outflow tract. Fibrous callus was resected by video assisted thoracic surgery [60].

Delayed pericardial events (n=5)

Moss and coworkers [27] reported a case of major sepsis: bilateral pleural empyema and purulent pericarditis requiring pericardiectomy in addition to the Nuss-bar removal.

In a series of 335 patients, Park and coworkers [28] reported a case of constrictive pericarditis, as a result of post-operative pericardial effusion, which required a pericardiectomy during Nuss-bar removal.

Finally, we identified three cases of late-onset pericardial tamponade. One patient was treated with pericardiocentesis under echocardiographic control [61]. Retrocession of the effusion was achieved after Nuss-bar removal in the second patient [62]. The third patient underwent pericardiocentesis, and then pericardial window for recurrence, associated with Nuss-bar removal [63].

Table 2. Characteristics and outcomes for 27 patients who sustained a procedural cardiac injury during MIRPE

Authors	Patient age (y) Prior surgery	Cardiac injury. (PCA)	Approach. (CPB)	PEx repair	Outcomes
Nuss[11]*, Hebra [26]*, Moss [27]*	(8)	Right atrium/ventricle	Sternotomy (CPB)	Ravitch	Recovery
Willekes [25]	(NM) « Child ».	Right atrium/ventricle, tricuspid valve	Sternotomy (CPB)	Ravitch	Recovery
Park [28]	(26) MIRPE, bar dislocation	Right atrium/ventricle (during Nuss-bar replacement)	Thoracotomy	Nuss-bar	NM
Gips [29]	(17)	Right and left atrium, Right ventricle. (PCA)	Thoracotomy (CPB)	Ravitch	Dead on Day 1
Belcher [30]	(16)	Right atrium	Sternotomy	Nuss-bar	Recovery
Castellani [32]	(NM) « Child ».	Right atrium/ventricle	Thoracotomy	NM	Recovery
Kuenzler [33]**, Bouchard (Case 3) [34]**	(11). Atrial septal defect closure	Right ventricle	Sternotomy (CPB)	none	Recovery
Bouchard (Case 1) [34]	(14)	Right atrium, ventricular septum	Sternotomy	none	Persistent ventricular septal defect
Bouchard (Case 2) [34]	(18) Ravitch-type repair	Right atrium/ventricle. (PCA)	Thoracotomy (CPB)	none	Severe hypoxic brain injury
Hernandez (Case 2) [13]	(5) Diaphragm hernia repair	« Cardiac perforation »	NM	none	Recovery. Nuss-bar under CPB at 11
Tedde [36]	(NM)	« Cardiac perforation »	NM	NM	Recovery
Becmeur [35]	(18)	Right atrium/ventricle	Sternotomy (CPB)	none	Recovery
Schaarschmidt [37]	(16)	Right atrium/ventricle (PCA)	Sternotomy	none	Dead on Day 11
Umuroglu [38]	(NM)	« Myocardic laceration »	NM	Nuss-bar	NM
Craner [39], Jaroszewski [46] **	(29) Mitral valve repair	Right atrium	Sternotomy (CPB)	Nuss-bar	Recovery
Jeong [40]	(17)	Left ventricle laceration	Sternotomy	Nuss-bar	Recovey
Zhang [41]	(11) Ventricular septal defect closure	Right atrium (PCA)	NM	NM	Dead on Day 17
Pilegaard [43]	(NM). MIRPE, recurrence	Right ventricle laceration	Sternotomy	Nuss-bar	Recovery
Ersen [42]	(NM) « Adult »	« Small ventricle defect »	Thoracotomy	Nuss-bar	Recovery
Li [45]	(NM) « Child » « Cardiac surgery »	« Cardiac perforation »	NM	NM	NM
Jaroszewski (Park's case) [46]	(36) Mitral valve replacement	Right atrium	Sternotomy (CPB)	Nuss-bar	Recovery
Jaroszewski (Obemeyer's case) [46]	(20) Transposition of great vessels	Right atrium	Sternotomy (CPB)	Nuss-bar	Recovery
Jaroszewski (Obemeyer's case) [46]	(NM) Atrial/ventricular septal defect closure	Right atrial appendage	Sternotomy	Nuss-bar	Recovery
Jaroszewski (Yüksel's case) [46]	(22) Atrial septal defect closure	Right atrium	Thoracotomy.	Nuss-bar	Recovery
Ghionzoli [44]	(14)	Right atrium	Thoracotomy	Nuss-bar	Recovery
Chen [47]	(NM) Atrial septal defect closure	Right atrium	Sternotomy	Nuss-bar	Recovery
Chen [47]	(NM) Atrial septal defect closure	Right atrium	Sternotomy	Nuss-bar	Recovery

*: threefold published case; **: twofold published case; NM: Not Mentioned; PCA: Procedural Cardiac Arrest; CPB: Cardiopulmonary By-Pass

MIRPE and cardiac arrest

Severe procedural complications, either during the initial operation or later Nuss-bar removal, were responsible for cardiac arrest in eight patients [29,34,41,52,54,57]. Three of them died early and three after a long coma; and one retained severe hypoxic brain injury [34]. Finally, only one patient fully recovered [54].

Redlinger and coworkers reported two intraoperative cardiac arrests in their series of 100 cases of re-intervention by MIRPE for PEx recurrence, in relation to right ventricular dysfunction in patients having undergone a previous Ravitch procedure and consecutive acquired thoracic dystrophy. Cardiac arrest occurred at the beginning of re-intervention by MIRPE, and Nuss-bar removal, respectively [64]. Recently, a case of unintended intraoperative cardiac arrests was reported by Zou and coworkers: two attempts for intrathoracic placement of the Nuss-bar resulted in cardiac arrest, which reverted after Nuss-bar removal. The PEx cure was therefore abandoned [65].

Late-onset cardiac arrest, probably as a consequence of cardiac rhythm disorder, is a tragedy because the presence of the Nuss-bar prohibits effective cardiopulmonary resuscitation manoeuvers as demonstrated by three well-documented cases [66-68]. Finally, two additional lethal late-onset cardiac arrest in patients with the Nuss-bar in place were reported in literature, however without any information about the cardiopulmonary resuscitation manoeuvers that might have been implemented [2,69].

In total, among the eight patients who sustained a procedural or late-onset cardiac arrest without any surgical complications, five died and one suffered hypoxic brain injury [64].

Comment

The present study is an overview of severe adverse events involving the heart and/or large mediastinal vessels occurring during PEx surgery or later. In the field of open repair, we identified 13 LCAEs resulting in one death, and 56 related to MIRPE resulting in 11 deaths, and two cases of severe hypoxic brain injury as a consequence of intraoperative cardiac arrest.

In open surgery, the use of Kirchner wire for fixation of the remodeled anterior chest wall is currently abandoned, and the reported postoperative cardiac perforation [14] should no longer be observed. Other LCAEs were the result of technical errors and/or a failure to follow up patients over the mid/long term, thus impeding to remove the metallic material in a timely manner. Therefore, it has been shown that intrapericardial or intracardiac migration of metallic materials used in PEx open repair was related to the time these materials were left in place, or as a consequence of delayed material rupture [19]. Intracardiac migrations of the sternal support bar should be avoided by using material of sufficient length, thus avoiding unintended dislocation into the mediastinum; and rigid enough to avoid torsion or rupture [8]. Another basic precaution is removing the bar at six months after PEx repair. Finally, the use of steel wire for sternal osteotomy fixation should be avoided because of the risk of rupture and subsequent intrapericardial or even aortic migration of the broken material [24].

The LCAEs related to MIRPE which we identified are much more worrying by their nature and number. In this regard, in Kenzler and Stolar's general review of PEx surgical management it is significant that the paragraph on surgical complications deals primarily with MIRPE [33]. A recent article by Hebra and coworkers [70] is expected to be an overview of miscellaneous severe adverse events and mortality related to MIRPE. Surprisingly, they identified few LCAEs in the literature: 19 cases, including 12 cardiac perforations and four cases of mortality. Furthermore, in their article, the crucial problem of cardiac arrest during MIRPE or later was not mentioned. Despite the fact that Hebra and coworkers pooled the miscellaneous severe adverse events that they identified with unpublished cases from a survey of members of the Chest Wall International Group, the morbidity and mortality of MIRPE remained severely underestimated, which we stressed in a correspondence [71]. In fact, by pooling the results of our present study (56 LCAEs) with the 20 unpublished LCAEs of the Chest Wall International Group' survey reported by Hebra and coworkers [70] the number of LCAEs related to MIRPE rises to 76, including 45 cardiac injuries, 18 cases of mortality and two cases of severe hypoxic brain injury. Notably, it has been shown that a history of median sternotomy for cardiac surgery was a significant risk factor for the occurrence of cardiac injury with a reported rate from 4 to 7% [46,47]. Thus, we feel that MIRPE might be discouraged in the setting of PEx repair in this patient population [72].

We are convinced that the technique refinements developed over the years to avoid cardiac injuries, well described in detail by Becmeur and coworkers [35], and then by Hebra and coworkers [70], have undoubtedly helped to reduce the number of LCAEs related to MIRPE. However, some of them are unpredictable, such as acute obstructions of the inferior vena cava, procedural cardiac arrests, or delayed cataclysmic hemorrhages especially during later Nuss-bar removal. Last, the impossibility of implementing effective cardiac resuscitation manoeuvers due to the presence of the Nuss-bar in the event of late-onset cardiac arrest deserves to be stressed [67].

Finally, whatever the PEx repair performed, the occurrence of a LCAE frequently mandates a cardiac/aortic procedure under CPB. After open repair, there were scheduled procedures in the majority of cases (6/7). In contrast, after MIRPE, these CPB procedures were mainly performed in an emergency setting (10/12). Thus, for safety reason, MIRPE should be carried out in an operating room where a bypass team can be brought in if necessary [34].

We are well aware of the imperfect nature of our study, since the number of patients, operated by open repair or MIRPE, respectively, is unknown. Thus, it seems questionable to extrapolate from a rate of severe or lethal complications after MIRPE as Hebra and coworkers did in their study [70]. However, given their actual number, their severity and the unintended nature of such complications, we did not include MIRPE in our therapeutic armamentarium for the treatment of PEx. When the operation is considered we remain faithful to the use of a simplified Ravitch-type repair [7], which has been demonstrated as safe and reliable, notably in patients with a past history of cardiac surgery [72,73].

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