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Pectus excavatum repair after sternotomy: the Chest Wall International Group experience with substernal Nuss bars

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Abstract

OBJECTIVES: Patients with pectus excavatum (PE) after prior sternotomy for cardiac surgery present unique challenges for repair of PE. Open repairs have been recommended because of concerns about sternal adhesions and cardiac injury. We report a multi-institutional experience with repair utilizing substernal Nuss bars in this patient population.

METHODS: Surgeons from the Chest Wall International Group were queried for experience and retrospective data on PE repair using substernal Nuss bars in patients with a history of median sternotomy for cardiac surgery (November 2000 to August 2015). A descriptive analysis was performed.

RESULTS: Data for 75 patients were available from 14 centres. The median age at PE repair was 9.5 years (interquartile range 10.9), and the median Haller index was 3.9 (interquartile range 1.43); 56% of the patients were men. The median time to PE repair was 6.4 years (interquartile range 7.886) after prior cardiac surgery. Twelve patients (16%) required re-sternotomy before support bar placement: 7 pre-emptively and 5 emergently. Sternal elevation before bar placement was used in 34 patients (45%) and thoracoscopy in 67 patients (89%). Standby with cardiopulmonary bypass was available at 9 centres (64%). Inadvertent cardiac injury occurred in 5 cases (7%) without mortality.

CONCLUSIONS: Over a broad range of institutions, substernal Nuss bars were used in PE repair for patients with a history of sternotomy for cardiac surgery. Several technique modifications were reported and may have facilitated repair. Cardiac injury occurred in 7% of cases, and appropriate resources should be available in the event of complications. Prophylactic re-sternotomy was reported at a minority of centres.

Keywords: Pectus excavatum • Revision surgery • Sternotomy • Minimally invasive surgery • Nuss • Complications

INTRODUCTION

Pectus excavatum (PE) is a common chest wall malformation characterized by posterior depression of the sternum and adjacent costal cartilages [1–3]. Coexistent cardiac defects can be seen especially with connective tissue disorders such as Marfan familial syndrome [2, 3]. PE may also be more common in patients who underwent a sternotomy at a young age [4]. Repair of PE after sternotomy can be difficult because of substernal adhesions, chest wall rigidity and ossification [4–7]. Although Ravitch-type (open) repairs following previous sternotomy have been reported, few publications describe the role of substernal Nuss bars after sternotomy for cardiac surgery [4, 5]. The Nuss procedure, introduced in 1998, has become increasingly popular because it can be used to correct PE without cartilage resection and sternal osteotomy [8]. This approach may have advantages over open repair by limiting additional excisions and osteotomies. Placing substernal Nuss bars requires that the mediastinal space be accessed and in patients who have had a prior sternotomy for cardiac surgery, the pericardium or epicardium of the right heart may be adhered to the sternum [5–7]. The additional risks and potential complications of substernal Nuss bar placement in this patient population have not been reviewed. The experience of multiple institutions performing PE repair utilizing substernal Nuss bars after sternotomy and cardiac surgery is presented.

MATERIALS AND METHODS

An appeal at the annual Chest Wall International Group meeting was made, and a survey was sent to 450 surgeons representing 260 centres in 52 countries that are members of the organization. An initial email and 3 follow-up reminders were sent from January 2015 through December 2015. Member surgeons were asked to submit responses about all of their patients who had PE repair and who met the following criteria: (i) PE deformity with a history of sternotomy for cardiac surgery and (ii) an attempted PE repair with substernal Nuss bar placement. The study included patients with or without elective resternotomy, urgent sternotomy, sternal osteotomy or previous PE repair with cartilage excision of any type (prior Ravitch). Institutional review board approval was obtained from countries requiring such reporting, and individual patient consent was obtained when required. Results from patient clinical records, including demographic characteristics, operative course, outcomes and experience, were collected and summarized for patients operated on from November 2000 to August 2015 (S. Li, S. Tang, and L. Yang patients from February 2005 to November 2012). Preoperative indications varied by institution but most included a Haller index of 3.25 or greater, symptoms secondary to the deformity and/or significant psychological impact from the PE [1]. Follow-up included clinical visits and chest roentgenograms with bar removal recommended at 2–3 years.

Statistical analyses were performed using Microsoft Excel 2010 (Microsoft.com, Redmond, WA, USA). For data not normally distributed, the median and interquartile ranges (IQRs) are reported.

RESULTS

Data for 75 patients were available from 14 centres. In total, 41 responses were received; 27 (66%) centres reported no experience.

The median age at PE repair was 9.5 years (IQR 10.9); the median Haller index was 3.9 (IQR 1.43) and 56% of patients were men. The median time to PE repair was 6.4 years (IQR 7.9) after prior cardiac surgery. Age and timing to repair varied considerably by centre; therefore, a meaningful analysis of these variables was not possible. The patients' demographic characteristics are presented in Table 1.

Four centres ($n = 12$ patients) performed redo sternotomy before pectus support bars were placed for some or all of their cases (7 elective and 5 emergent for bleeding or cardiac injury). Subsequent substernal placement of pectus support bars was performed with the sternum open in 4 cases and thoracoscopically after closure of sternotomy in the remainder. Four patients had a history of a failed Ravitch procedure (3 for excavatum and 1 for carinatum repair). In one of these patients, an open excision of malunion and titanium plating were performed in addition to the placement of substernal pectus support bars [9]. Table 2 reviews the institutions' reported cases and outcomes.

The median blood loss was 10 ml (IQR 25 ml) but ranged from 10 ml to over 4700 ml in cases complicated by cardiac injury. Five cases (7%) of cardiac perforation were reported, and emergent femoral bypass was used for rescue in 2 of these cases (Table 3).

Most patients (73%) had 1 pectus bar placed. Two bars were used in 21% of patients, and 3 bars were used in 4% of patients (2 patients had an unreported number of bars). Data on length of the bars were not collected. Four centres (5 patients) used lateral stabilizers (right sided: 1 patient, bilateral: 4 patients), and 2 centres (5 patients) placed medial stabilizers [4, 8, 10–16]. For stabilization in the rest of the patients, multipoint bar fixation

Table 1: Demographic characteristics of 75 patients who underwent a modified Nuss for pectus excavatum repair after sternotomy

Variables	No. of patients/ variable (%)
Age (years) at PE repair [median (IQR)]	9.45 (10.9)
Gender, male	42 (56)
Haller index [median (IQR)]	3.9 (1.43)
Marfan familial syndrome	6 (8)
Williams syndrome	1 (2)
Down syndrome	1 (2)
Triple X syndrome	1 (2)
Prior pectus repair (open Ravitch-type):	4 (6)
1 carinatum and 3 excavatum	
Prior cardiac surgery [median (IQR)]	
Years from cardiac surgery to pectus repair	6.35 (7.9)
Primary cardiac diagnosis/repair ^a	
Ventricular septal defect	33 (44)
Atrial septal defect	21 (28)
Patent ductus arteriosus	6 (8)
Mitral valve repair	4 (6)
Transposition of great vessels	3 (4)
Tetralogy of Fallot	3 (4)
Total anomalous pulmonary venous return	2 (3)
Aortic valve replacement	2 (3)
Ascending aortic root aneurysm	1 (1)
Supra aortic stenosis/aortoplasty	1 (1)
Double-outlet right ventricle	1 (1)
Not reported	3 (4)

^aSome patients had multiple defects reported.

IQR: interquartile range; PE: pectus excavatum.

Table 2: Centres reporting substernal Nuss bar placement for repair of pectus excavatum after prior sternotomy and cardiac surgery

Surgeon, centre	Total cases	Procedure techniques	CPB available	Use of sternal elevation	Cardiac injury, rescue and associated EBL	Median hospital length of stay, median months follow-up and other major complications
S. Li, S-T Tang, L. Yang ^a , Union Hospital, Wuhan, China	30 ^a	Right thoracoscopy used 100% Assisted with subxiphoid blunt dissection	100%	20%	None reported	Hospital days: 6 2 patients, pneumothorax 1 patient, bar displacement non-operative 1 patient, wound haematoma All bars removed uneventfully Follow-up reported, median 69 months Outcomes reported: 25 excellent 3 good 2 fair
S. Uemura, Kawasaki Medical School, Kurashiki, Japan	11	Right thoracoscopy used 100% Assisted with subxiphoid blunt dissection	No	Sternal hook 100%	None reported	Hospital days: 11 1 patient thoracentesis haemothorax All bars removed uneventfully Last follow-up reported was bar removal at median 24 months
H.J. Park, Seoul St. Mary's Hospital, Seoul, South Korea	10	Blind approach (before era of pectoscope): 3 Pectoscopy used: 3 Assisted with subxiphoid direct vision sharp dissection Sternotomy: 4 pre-emptive 1 emergent	90%	90%	Right atrium CPB, sternotomy EBL: 4700 ml	Hospital days: 5 9 patients with bars removed uneventfully Follow-up: 115 months All patients reported good-to-excellent results
D.E. Jaroszewski, Mayo Clinic, Phoenix, AZ, USA	4	Sternotomy: 3 pre-emptive and 1 emergent Right thoracoscopy used 100%	100%	50%	Right atrium converted to CPB, sternotomy EBL: 3000 ml	Hospital days: 7 1 patient thoracentesis for pleural effusion 2 patients bars removed uneventful Follow-up: 37 months All patients reported excellent results
H. Pilegaard, Aarhus University Hospital, Skejby, Denmark	3	Right thoracoscopy used 100% Assisted with subxiphoid blunt dissection	No	No	None reported	Hospital days: 3 All bars removed uneventful Last follow-up was at bar removal median 37 months
J.R. De Campos, Hospital das Clinicas, Sao Paulo, Brazil	3	Bilateral thoracoscopy 100% Assisted with subxiphoid blunt dissection	66%	Hand-held retractor 33%	None reported	Hospital days: 5 1 patient thoracentesis for pleural effusion Bars in place Follow-up 34 months All patients reported excellent results
R. Obermeyer, F.W. Frantz, R.E. Kelly, Children's Hospital of the King's Daughters, Norfolk, VA, USA	3	Right thoracoscopy used 100% Assisted with subxiphoid blunt and sharp dissection sternotomy: 2 emergent	100%	66%	Right atrium, converted to sternotomy EBL: 2000 ml Right atrial appendage limited sternotomy EBL: 15 ml	Hospital days: 5 1 patient, bars removed uneventfully No long-term follow-up reported on 2 patients 1 patient last follow-up at 36 months bar removal, excellent results
M. Yüksel, Marmara University Hospital, Istanbul, Turkey	3	Right thoracoscopy used 100% Assisted with subxiphoid blunt dissection in 1 case	100%	33%	Right atrium conversion to right anterior thoracotomy EBL: 500 ml	Hospital days: 7 1 patient bar removed and carina-tum treated Follow-up 41 months on 2 patients, 1 patient lost to follow-up

Continued

Table 2: Continued

Surgeon, centre	Total cases	Procedure techniques	CPB available	Use of sternal elevation	Cardiac injury, rescue and associated EBL	Median hospital length of stay, median months follow-up and other major complications
M. Torre, Istituto G. Gaslini, Genoa, Italy	2	Right thoracoscopy used in 1 case subxiphoid blunt dissection used in 1 case	50%	No	None reported	Hospital days: 6 1 patient with bar removed uneventfully 1 patient reported deceased at 3 years of medical issues unrelated to PE repair Last follow-up at 36 months bar removal
F-M. Haecker, University Children's Hospital, University of Basel, Basel, Switzerland	2	Bilateral thoracoscopy used 100% Assisted with subxiphoid blunt dissection	No	No	None reported	Hospital days: 12 Postoperative pericarditis in 1 patient with subsequent cardiac laceration and bleeding during bar removal 1 patient bar removed uneventful Follow-up 59 months Patients satisfied with results
L. McMahon, D. Notrica, Phoenix Children's Hospital, Phoenix, AZ, USA	1	Right thoracoscopy used 100% Assisted with subxiphoid blunt dissection	100%	100%	None reported	Hospital days: 4 Carinatum formation requiring open resection of deformed cartilage 1 year postoperative Bars removed with follow-up: 47 months Satisfied with results after reoperation
A. Hebra, Medical University of South Carolina, Charleston, SC, USA	1	Right thoracoscopy used 100% Assisted with subxiphoid blunt dissection	No	No	None reported	Hospital days: not reported Bars removed uneventful Last follow-up at 36 months bar removal
C. Chu, Country Hospital, Taipei, Taiwan	1	Right thoracoscopy used 100% Assisted with subxiphoid blunt dissection	No	No	None reported	Hospital days: not reported Bar in place with follow-up: 32 months Excellent results
J.D. Phillips, WakMed Health and Hospitals, Raleigh, NC, USA	1	Right thoracoscopy used 100% Assisted with subxiphoid blunt dissection	100%	100%	None reported	Hospital days: 5 Bar in place with follow-up at 19 months Preoperative symptoms improved

EBL: estimated blood loss; CPB: cardiopulmonary bypass.

^aPatient data reported from November 2000–August 2015.

techniques were used with pericostal wire/absorbable suture, FiberWire [17], claw fixators, hinge plates or bridge stabilizers [18].

The median hospital stay was 6 days (IQR 2.8) and varied among institutions (Table 2). The 30-day postoperative complications reported are presented in Table 2.

Follow-up was heterogeneous and reported for 63 patients (84%) at a median of 56 months (IQR 34.5). No standardized measurement of outcomes was obtained. Table 2 lists the centres that had available follow-up and outcomes. Subsequent reoperation was required in 2 patients for acquired carinatum and in 1 patient for bar rotation. Information on bar removal was reported for 73 patients, with bars having been removed in 61 patients (81%). Ventricular laceration

with haemorrhage during attempted bar removal was reported for 1 patient [19] who also experienced postoperative pericarditis after pectus repair. This patient had a history of a Mustard procedure for transposition, without pericardial closure. The remainder of reported bar removals were uneventful. A single patient was reported to have died 3 years postoperatively from unrelated medical issues.

DISCUSSION

Subsequent repair of PE after prior sternotomy for cardiac surgery presents technical challenges [5–7]. Dense mediastinal

Table 3: Demographic characteristics and outcomes for 5 patients with cardiac injuries

Surgeon, centre	Patient age, gender	Previous cardiac surgery, details	Time from cardiac surgery to definitive PE procedure	Surgical technique for PE repair	Cardiac injury and long-term outcomes
H.J. Park, Seoul St Mary's Hospital, Seoul South Korea	36-year-old, male	Mitral valve replacement with CPB, pericardium was not reported closed	5 years	Pectoscope used for visualization and dissection of pathway through mediastinum, introducer and initial insertion of bar uneventful. CPB on call. Sternal crane-lift elevation	Right atrium torn with rotation of bar due to adhesion between sternum and heart. Sternotomy and femoral CPB initiated; primary repair of atrium performed. Transfusion of >10 units due to 4700 ml EBL. Bar removed without incidence
D.E. Jaroszewski, Mayo Clinic, Phoenix, AZ, USA	29-year-old, male, Marfan familial syndrome	Mitral valve repair with annuloplasty and maze on CPB, postoperative complications of pericardial effusion and pericardial window, complete heart block requiring pacemaker placement, pericardium was reported closed	17 years	Bilateral thoracoscopy with blunt dissection of mediastinum, CPB on call and no sternal elevation	Right atrium torn with mediastinal dissection due to direct adhesion of atrium to posterior sternum, femoral bypass initiated and sternotomy performed to free additional adhesions; primary repair of atrium, transfusion of 12 units for EBL 3000 ml. Bars removed at 3 years without complication
R. Obermeyer, F.W. Frantz, R.E. Kelly, Children's Hospital of the King's Daughters, Norfolk, VA, USA	20-year-old, male, not reported	Repair transposition of the great vessels on CPB, pericardium not closed, ASD and VSD repair on CPB. Unknown whether pericardium closed	17 years, not reported	Right thoracoscopy with blunt dissection of mediastinum with subxiphoid-assisted dissection. CPB on call. Sternal elevation Right thoracoscopy with direct visualization dissection of mediastinum with directly visualized sharp subxiphoid dissection. CPB on call	Right atrium torn with mediastinal dissection due to direct adhesion of atrium to posterior sternum. Sternotomy performed for primary repair of atrium. Transfusion not reported, units for EBL 2000 ml Right atrial appendage adhesion to sternum with minimal bleeding; mediastinal dissection completed with partial open sternotomy. No transfusion EBL 15 ml. Bars removed at 3 years without complication
M. Yüksel, Marmara University Hospital, Istanbul, Turkey	22-year-old, male	ASD closure with CPB, pericardium closed with biologic patch	11 years	Pectoscopy with blunt dissection of mediastinum	Right atrial laceration with blunt mediastinal dissection. Conversion to right anterior thoracotomy and oversewn under direct visualization. Transfusion given, unknown units with estimated 500 ml of blood loss. Bars remain in place

ASD: atrial septal defect; CPB: cardiopulmonary bypass; PE: pectus excavatum; VSD: ventricular septal defect.

adhesions between the sternum and heart or great vessels may potentially lead to injury during reoperation and attempted dissection [5–7, 20, 21]. Few publications describe the associated risks, results and techniques of staged PE repair after sternotomy and cardiac surgery [4–7]. Some authors have recommended open, Ravitch-type procedures for this patient population [2, 6, 7, 22]; cardiac injury has not been reported for the Ravitch procedure, although little has been published about this. The risk likely

correlates with the extent of mediastinal dissection and sternal mobilization performed if a posterior strut is placed. The use of Nuss-type procedures, including for more complex procedures, continues to increase [4, 5, 12, 15, 23–25]. Therefore, understanding the potential risks in this patient group is critical: our series identifies additional risks of cardiac injury (7%) during repair with substernal bar placement and during bar removal (2%). By comparison, intraoperative adverse events in patients undergoing

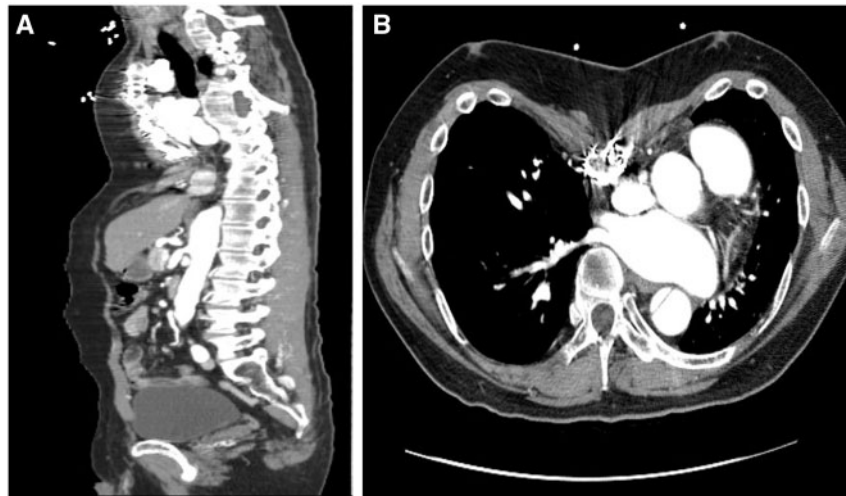


Figure 1: (A, B) Computer tomographic imaging of patients with prior sternotomy and pectus excavatum can be useful for assessment of anatomical relationships and surgical planning.

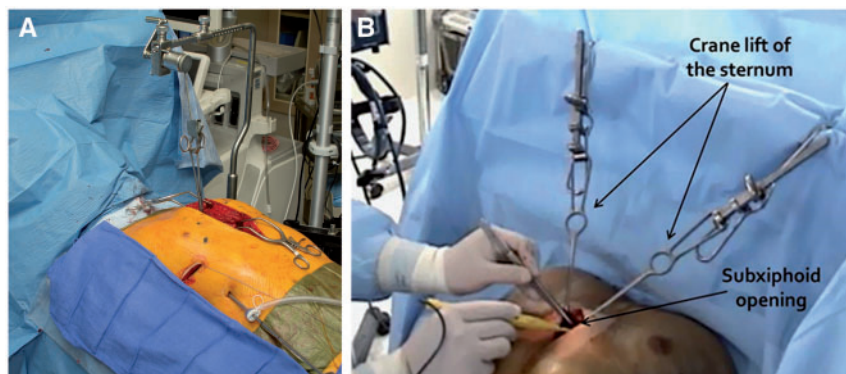


Figure 2: Use of forced sternal elevation allows elevation of the defect and may assist in direct visualization both across the mediastinum and from a subxiphoid approach for dissection. (A) Rultract elevation [29] and (B) crane-lift elevation [16].

reoperative cardiac surgery are also reported for up to 7% of patients, with most events involving dissection of mediastinal adhesions [20, 26]. Catastrophic haemorrhage during chest re-entry after prior sternotomy for cardiac surgery is a potential complication, and adequate preparation for control of bleeding, cardiotomy repair and resuscitation is imperative. For these cases, a cardiothoracic surgeon with immediate availability of a cardiopulmonary bypass team should be considered. Extensive informed patient consent should take place reviewing the substantial risks for reoperation after prior sternotomy and cardiac surgery.

Age and timing to repair varied considerably by centre, and meaningful analysis or recommendation of a timeline that would decrease the risk for repair was not possible. Prior operative notes and information about postoperative complications were obtained, when possible. There were no demographic differences or additional risk factors identified among the 5 patients who experienced cardiac injury nor was there an association by type of prior cardiac procedure. Publications about resternotomy for indications other than pectus repair have noted an increased risk associated with aortic root replacements, use of synthetic graft material, prior ventricular support, incomplete or missing pericardium and multiple sternotomies [5, 20, 21, 27, 28]. Postoperative complications, including pericarditis, mediastinal

haematoma and infection, may also increase risks [7]. A patient who had an injury at the time of bar removal had experienced postoperative pericarditis, which may have contributed to excessive adhesion formation [19]. Preoperative evaluation with computerized tomographic imaging has been helpful in reoperative cardiac cases to visualize disturbed anatomic relationships of the heart and great vessels with the sternum [20, 27] (Fig. 1A and B). In prior studies by Roselli *et al.* [20] of patients undergoing reoperative sternotomy, the lack of preparative imaging was identified as the most common missing element associated with injury.

Techniques used for dissection of the mediastinum may additionally influence risks. In two-thirds of the cases with inadvertent cardiotomy, right atrial injury occurred during blunt mediastinal dissection. All of these cases were performed with thoracoscopic direct visualization and blunt dissection across the mediastinum, which did not prevent injury. Even with the sternum cleared, remaining adhesions may cause bleeding. Right atrial tearing was noted with bar rotation and resulted from mediastinal adhesions that had not been completely cleared. A subxiphoid approach for assistance in the takedown of mediastinal adhesions was used by multiple centres. Blind dissection is highly discouraged by cardiac surgeons experienced with reoperative sternotomy, and a significant risk is associated with subxiphoid blind, blunt-finger dissection [28]. Although not included in this



Figure 3: Patients should be positioned supine with the groin prepared in a surgical field should emergent access be necessary by sternotomy and groin vessels.



Figure 4: If sternotomy is performed, the edges of the sternum may have to be recut at an angle to allow approximation after elevation of the excavatum defect.

review, Li *et al.* [5] reported a cardiac perforation during blind, blunt dissection through a subxiphoid incision and have since modified their technique using sharp dissection from a subxiphoid access and thoracoscopy for visualization from the right side.

In the authors' opinion, when dense adhesions under the sternum are present or the epicardium is suspected to be adhered, a complete or partial reopening of the previous sternotomy with an oscillating saw should be performed. Direct vision resternotomy is a common technique employed by cardiac surgeons when only adhesions directly visualized from the subxiphoid view are divided and when only the sternal bone that has been separated from adhesions is divided [28]. An elective (pre-emptive) resternotomy was performed by 2 centres in cases where the epicardium was suspected to be adhered to the sternum. Rapid conversion to partial sternotomy also minimized blood loss and prevented further injury at another centre, where adhesions of the atrial appendage to the sternum were identified. Use of assisted sternal elevation (Fig. 2A and B) or other sternal lift procedures may be helpful to create working space between the depressed sternum and the pericardium [15, 16, 29, 30]. Most centres (64%) used some type of assisted sternal lift for some or all of their cases. Although there is no direct data to support a decrease in risk with elevation of the sternum, increased visualization is desirable.

Both the surgeon and the operating room team should expect a significant risk of cardiac injury during repair and a small but significant risk at bar removal. A well-coordinated rescue strategy should be reviewed with the operative team, and the expected action should be ready to execute should an injury occur [27, 28]. Both femoral areas should be prepared and draped into the surgical field (Fig. 3). In high-risk patients, an existing femoral arterial line can facilitate emergent percutaneous arterial inflow cannulation for cardiopulmonary bypass, if needed [27, 28]. We recommend that a primed bypass pump, blood and sternal re-entry saw be immediately available for high-risk cases. Although only 2 cases that required emergent bypass were reported, the risk of cardiac injury supports this resource. If a cardiac injury occurs or substantial bleeding is encountered, expeditious initiation of rescue manoeuvres must be made by the operating surgeon and/or cardiac surgeon. The chest should be packed to slow blood loss during sternotomy. If cardiac injury is suspected during instrument passage, the instrument should not be removed to help maintain tamponade until the injury is accessed via sternotomy.

If the pericardium is not intact or a substantial pericardiotomy occurs with mediastinal dissection, closure of the sac should be done primarily or with a graft to attenuate the risks of adhesion formation between the bar and epicardium. If a reoperative sternotomy is performed, closure of the sternum may require remodelling of the sternal edges to properly approximate [24] (Fig. 4). Plating of the sternum can also be considered to reinforce integrity.

Limitations

This study was subject to all the limitations of a retrospective review. Additionally, most of the centres had relatively small experiences to contribute, and the non-response rate was high. This was a non-random sample, and the data were not normally distributed. There was substantial variability in techniques, documentation and outcomes observed among the institutions from many countries. The study was subject to sample bias and to strong potential biases of centres not wanting to report their adverse data. No consistent objective or even subjective measurements after surgery were available for this heterogeneous population. Our intention was to review the wide breadth of experience available and to provide recommendations in an area where limited data currently exist.

CONCLUSION

A broad range of institutions used substernal Nuss bars to repair PE in patients with prior sternotomy for cardiac surgery. The risk of cardiac injury was greater in this population; therefore, informed consent should be extensive, and patients should understand the potential catastrophic complications that may occur. Surgeons should anticipate potential cardiac injury and have available appropriate resources for repair. Elective resternotomy was used for a number of cases and may be considered for patients with substantial mediastinal adhesions and for cases where there is concern for epicardial sternal adhesions. Several technique modifications were reported from centres that may have facilitated repair.

Conflict of interest: none declared.

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