One-step surgery for cyanotic heart disease with pectus excavatum: Should it be done?

Yuletta Adny Ambarsari, PhD[1], Yunanto Kurnia, MD[1], Haryo Aribowo, MD[1], Ihsanul Amal, MD[1], Supomo Supomo, MD[1], Trianingsih Trianingsih, MD[2], Tandean Tommy Novenanto, MD[3], Dicky Panditatwa Susantya, MD[3]

[1]Division of Cardiothoracic and Vascular Surgery, Department of Surgery, Faculty of Medicine, Public Health and Nursing, Universitas Gadjah Mada, Dr. Sardjito General Hospital, Yogyakarta, Indonesia, [2]Department of Radiology, Faculty of Medicine, Public Health and Nursing, Universitas Gadjah Mada, Dr. Sardjito General Hospital, Yogyakarta, Indonesia, 3Faculty of Medicine, Public Health and Nursing, Universitas Gadjah Mada, Yogyakarta, Indonesia

SUMMARY

Congenital heart disease accompanied with pectus excavatum is very rare, we present our experienced in one step surgery of pectus and cardiac repair and staged procedure at our institution. We retrospectively reviewed medical records for patients who underwent a hybrid repair of both pectus and CHD between 2022 and 2023 in RSUP dr. Sardjito, Indonesia. Of these patients, 2 patients had both pectus and CHD. The first patient was using one-staged produce operation began with ASD repair than the patient proceeded with pectus repair. During pectus repair, the patient experienced worsening hemodynamics, the total operation took time seven hours. During those seven hours, there was 1400 cc of bleeding. The patient's condition was stationary and worsened within 72 hours. The patient died three days after surgery due to sepsis, MODS, and hyperlactatemia. Meanwhile the second patient underwent two-staged produce showed good result. The total duration of the operation is half shorter than the one-step operation experience, bleeding during the operation also appears to be at least 300 cc. After surgery, the patient's condition was stable and the vital sign was satisfactory. The patient currently has no complaints, including no concerns about unstable hemodynamics caused by the pectus condition. The timing and approach to surgical correction of pectus excavatum in patients with congenital heart disease must be individualized. Factors such as age, anatomical development, cardiopulmonary status, and the complexity of the cardiac defect should guide the decision between single-stage and two-stage procedures. While early intervention carries the risk of recurrence due to ongoing growth, delayed or staged surgery may offer better long-term stability and outcomes, particularly in complex or adult cases.

KEYWORDS:
Pectus Excavatum, Congenital Heart Disease, One-stage repair, Two-stage repair, outcome

INTRODUCTION

Pectus excavatum (PE) is the most common congenital chest wall deformity, seen in up to 1 in 300–1,000 newborns, and predominantly affects males.[1] Meanwhile, congenital heart disease (CHD) affects about 9.4 per 1,000 live births, with acyanotic types like ASD, VSD, and PDA comprising the majority (57.9%) and cyanotic types such as TOF and TGA making up 8.2%.[2] Although both conditions are relatively common on their own, the coexistence of PE and CHD is rare. For instance, only 0.5% of CHD patients undergo PE repair, according to Hasegawa et al.[3] and just 0.17% in a major Boston study.[4]

Management of pectus excavatum (PE) includes a spectrum of approaches—ranging from pain control and conservative monitoring to surgical correction. Surgery is especially recommended for patients with severe chest wall deformities (Haller Index > 3.25), cardiopulmonary impairment, significant cosmetic concerns, or other related symptoms.[5-7] Two widely used surgical techniques include: The Ravitch procedure, a more invasive method that involves resecting the anterior costal cartilage and placing a mesh to support the chest wall.[8] The Nuss procedure, a minimally invasive technique in which a curved metal bar is inserted beneath the sternum to elevate the chest, typically left in place for 2 to 3 years.[9]

Historically, PE and CHD were treated through separate surgical procedures due to concerns over heightened risks of bleeding, infection, and anesthesia-related complications. However, recent studies have demonstrated the safety and success of combined surgical repairs, shifting this traditional paradigm.[10]

Despite these advancements, performing simultaneous repairs remains challenging. Procedures such as pectoral muscle flap elevation, costal cartilage resection, and sternal osteotomy—when combined with cardiopulmonary bypass (CPB) during CHD correction—can significantly increase the risk of intraoperative bleeding. Thus, although feasible, concomitant repair of PE and CHD requires careful planning and surgical expertise.[10]

Therefore, as a tertiary referral center in a developing country, we present our experience in the surgical management of patients with coexisting pectus excavatum

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Corresponding Author: Yuletta Adny Ambarsari
Email: aini1706@yahoo.com

and congenital heart disease. We performed single-stage repair in patients with acyanotic CHD and two-stage repair in those with cyanotic CHD, providing valuable insight into the safe and effective treatment of this rare and complex dual pathology.

CASE PRESENTATION

This case series features two female patients who have been diagnosed with congenital heart disease, and are scheduled for repair procedures to correct their cardiac anomalies. Notably, these patients exhibit the comorbidity of pectus excavatum, making them eligible for pectus correction. The study, conducted with ethical clearance number KE/FK/0256/EC/2024 from the Ethics Committee of the Faculty of Medicine, Public Health, and Nursing at Universitas Gadjah Mada, is a retrospective case series that includes a thorough examination of medical records for patients who underwent a hybrid repair of both pectus and CHD between 2022 and 2023.

Case 1

A 24-year-old female presents with a major complaint of shortness of breath, diagnosed with an atrial septal defect since the age of three months. Despite being urged to have heart surgery at the National Cardiovascular Center Harapan Kita Hospital at the time, she has not yet agreed to the procedure. Over the past year, she has had regular shortness of breath, fainting during intensive activities, and impaired growth and development. She was referred to our tertiary hospital after experiencing severe symptoms and is now receiving propranolol 3x10mg and furosemide 1x40mg. She denies a history of chest pain, cold sweats, swollen feet, heart attacks, heart failure, hypertension, diabetes, dyslipidemia, or family history. On examination, she is conscious and in good general condition, with normal vital signs. Physical examination reveals cardiomegaly, a wide fixed split S2 sound, and a continuous murmur in the sub valvular area. The laboratory results are normal, and chest X-ray findings confirm cardiomegaly and reveal right diaphragmatic eventration. Transthoracic echocardiography (TTE) reveals bidirectional shunt patent ductus arteriosus (PDA) with a diameter of 0.9-1.6cm, multiple left-to-right atrial septal defects (ASD) with diameters of 8mm and 6mm, severe pulmonary stenosis (PS), mild pulmonary regurgitation (PR), and mild tricuspid regurgitation (TR). Right heart catheterization (RHC) and contrast-enhanced cardiology computed tomography (CT) scan further confirm the echocardiography findings, which include no PDA, multiple left-to-right ASD with high flow (largest size 1.3cm), severe valvular PS, mild infundibular PS, proximal left pulmonary artery stenosis, dilated right atrium, and hypertrophic right ventricle. A thoracic CT scan also reveals pectus excavatum with a Haller index of 3.86. The patient undergoes corrective procedures, including ASD closure with transannular patch placement (cribriform ASD size 1cm x 0.5cm), correction of pulmonary stenosis with commissurotomy on fused cusps, widening of Right Ventricular Outflow Tract (RVOT), infundibulectomy, and reconstruction of pectus excavatum by cutting 1 cm of costal cartilage IX-XI and a modified Ravitch procedure with reconstruction plates and miniplates. Postoperative TTE shows an ejection fraction (EF) of 60%, Tricuspid Annular Plane Systolic Excursion (TAPSE) of 8mm, and a residual shunt of 0.8mm. The operation began with ASD repair, after weaning the first CPB with a duration of 88 minutes and Aox 57 minutes, the patient proceeded with pectus repair. During pectus repair, the patient experienced worsening hemodynamics, necessitating a repeat CPB with a duration of 23 minutes and Aox 21 minutes. The total operation took time seven hours. During those seven hours, there was 1400 cc of bleeding. After surgery, the patient was treated in the ICU during the post-operative period. The patient's condition was worsened within 72 hours. The laboratorium showed the finding of leukocytes up to 20.1, procalcitonin 7.26, and a trend of decreasing pH to 7.195 and increasing lactate from 2.9 up to 10.7 (hyperlactatemia). The patient underwent culture tracing, which did not grow germs in blood, phlegm, and urine. The urine results showing gross hematuria and albuminuria. The chest x-ray showed pneumonia, the patient's condition worsened after the vital sign was not improving with support. The patient died three days after surgery due to sepsis, MODS, and hyperlactatemia. The patient was hospitalized for five days.

Case 2

An 18-year-old female patient presented with a progressive onset of shortness of breath and fatigue, particularly during vigorous activities, over the past year. She reported using two pillows for sleeping daily but denied any interruption throughout the night. She was diagnosed with a congenital heart defect at the age of 7 months and reported cyanosis in her lips and nails, as well as shortness of breath and fatigue during excessive exercise. She had been treated at our tertiary hospital since she was toddler, and was taking propranolol 3x10 mg on a regular basis. As a student, she frequently tired during intensive activities but she denied episodes of coughing up blood, fainting, or chest pain in the past year, and she had not been hospitalized. She denies any history of diabetes, hyperthyroidism, hypertension, dyslipidemia, or a family history of similar conditions.

When the patient arrived at the emergency department complaining of shortness of breath, she was conscious and had normal vital signs except for an oxygen saturation of 80%. Physical examination revealed symmetrical chest movement, vesicular breath sounds, and the presence of pulmonary and mitral systolic murmurs with signs of cardiomegaly; clubbing of the fingers was also observed. Further evaluation through transthoracic echocardiography and right heart catheterization revealed a large bidirectional right-to-left shunt ventricular septal defect (VSD) measuring 1.2cm, along with a muscular VSD measuring 3cm. Hypertrophy of the right ventricle, overriding aorta, severe infundibular pulmonary stenosis, and the presence of PDA were also identified. Additional contrast-enhanced heart CT confirmed Tetralogy of Fallot (TOF) with misaligned VSD and pulmonary stenosis with collaterals. A thorax CT scan revealed the presence of pectus excavatum with a Haller index of 3.55.

The patient was scheduled for corrective surgery to treat the malalignment, which included TOF correction with VSD closure via a pericardial patch and infundibulecomy for

Table I: Surgical Data Comparison of Case 1 and 2

	Case 1	Case 2
Surgical management	Atrial septal defect closure with Ravitch procedure	Total correction without Pectus Excavatum repair
Aortic cross clamp time (min)	57 + 21	99
Cardiopulmonary bypass time (min)	88 + 23	124
Surgery time (min)	435	210
Intraoperative bleeding (cc)	1400cc	300cc

min: minute, cc: cubic centimeter

Patent Foramen Ovale (PFO) closure. Despite the high Haller index, no correction for pectus excavatum was undertaken. Postoperative TTE demonstrated improvement in both left and right ventricular function, with Left Ventricular Outflow Tract Diameter (LVOTd) measuring 15mm, Left Ventricular Outflow Tract Velocity Time Integral (LVOTVTi) measuring 14.6mm, Left Ventricular Internal Diastolic Diameter (LVIDd) measuring 30mm, EF of 70%, and TAPSE of 10mm. Based on experience of doing one step surgery resulting with the long duration of surgery and deceased patient outcome. This time, the patient was decided to undergo two step surgery which TOF repair was carried out first. Apart from the condition of the patient's haler index which was lower than the experience of the first patient, the patient underwent total correction surgery with CPB time 124 minutes and aox time 99 minutes. The total duration of the operation is half shorter than the one-step operation experience, bleeding during the operation also appears to be at least 300 cc. After surgery, the patient's condition was stable and the vital sign was satisfactory; laboratory evaluation revealed a modest rise in leukocytes up to 12.2 with normal pH and lactate 2.9; the patient spent 30 hours in the ICU and 23 hours on a ventilator. Patient was in the hospital for 8 days. Patient follow-up has been ongoing for up to 9 months following surgery. The patient currently has no complaints, including no concerns about unstable hemodynamics caused by the pectus condition. Comparative data from Case 1 and Case 2 are presented in Table I.

DISCUSSION

Correction of pectus excavatum is recommended when patients exhibit high Haller index and correction index values. Additionally, indications for surgery include excessive pain, cardiopulmonary dysfunction, exercise intolerance, and cosmetic concerns.[7] Patients are often encouraged to have at least in the mid-adolescent age range, when they have likely reached their maximum growth, to minimize the risk of recurrence.[7] Pectus surgery can be performed in both adult and pediatric populations, with several factor influencing the patient's clinical improvement benefit.1[1] Both of our patients are late adolescent, who match the repair requirement for their PE condition. However, both patients are females, and PE in females is less prevalent than in males.[1] Even more unusual, both patients have congenital heart disease, one with left-to-right shunt atrial septal defect (ASD) with pulmonary stenosis (PS), and the other with Tetralogy of Fallot (TOF). According to Liu et al., ASD account for 15.3% of all congenital heart disease (CHD) cases, whereas TOF has a prevalence of 4.4%.[2] Managing patients with both CHD and PE is challenging due to the rarity of such cases.

Several studies have reported extensive experience with combined surgical correction of cardiac defects and pectus deformities. In these reports, simultaneous procedures were commonly performed in patients with acyanotic congenital heart disease and in individuals with Marfan syndrome, yielding promising outcomes. However, for more complex cardiac anomalies, including tetralogy of Fallot (TOF), transposition of the great arteries (TGA), double outlet right ventricle (DORV), complete atrioventricular canal (CAVC), and hypoplastic right ventricle—a staged approach was preferred, with intracardiac repair performed prior to pectus correction to minimize surgical risk and optimize patient stability.[3,12-13]

A one-stage procedure is regarded safer since correcting pectus excavatum first might have detrimental effect on wound healing, particularly in patients with severe cyanosis. Additionally, if the pectus is not addressed, the compression might cause serious hemodynamic compromise. In contrast, if the cardiac lesion is repaired but the pectus is not treated, compression from the pectus might disrupt postoperative hemodynamics.[3] The advantages of simultaneous surgery include only one general anesthetic and a single hospital stay.[13] It also requires optimal operative exposure during the cardiac procedure while minimizing bleeding risk, as well as optimal chest-wall stabilization postoperative. Furthermore, concomitant pectus repair may be recommended because it improves cardiovascular function by facilitating heart filling, potentially improving postoperative outcomes.[3] Additionally, significant increases in right ventricular end-diastolic volume index (RVEDVI) and right ventricular stroke volume index (RVESVI) have been observed. However, the disadvantages of a one-stage procedure include longer surgical time, more blood loss, and inadequate exposure to the heart.[3]

Therefore, in the first case, we tried to perform one step concomitant surgery by starting with ASD correction and continuing with pectus correction. Considering that one step procedure currently shows more promising outcomes and are already recommended for patients with acyanotic CHD, we opted for a one-stage surgical approach. This decision was based on the generally favorable prognosis of acyanotic CHD and the fact that corrective open-heart surgery in these patients typically does not require prolonged CPB. Our intraoperative findings supported this approach, as the patient remained hemodynamically stable following atrial septal defect (ASD) repair. Based on our first patient experience, prolonged operative time and significant intraoperative bleeding led to postoperative instability, ultimately resulting in serious complications, including sepsis, multiple organ dysfunction syndrome (MODS), and hyperlactatemia.[14] These adverse outcomes were primarily

attributed to several factor such as excessive blood loss and prolonged CPB duration. Excessive blood loss, particularly during cardiac surgery involving CPB, may result in anemia, hypovolemia, and impaired tissue oxygenation. These conditions weaken immune function and increase the risk of infection and sepsis. Prolonged CPB duration is associated with systemic inflammatory response activation and disruption of immune homeostasis, further predisposing patients to sepsis. CPB also contributes to hemodilution and depletion of clotting factors, increasing bleeding risk and the likelihood of transfusion, both of which heighten infection susceptibility.[15]

Additionally, technical challenges such as limited exposure and difficulty in optimal placement of the sternal retractor are frequently encountered during simultaneous cardiac and chest wall corrective procedures, further complicating the surgical field. Some authors experiences stated that a vertical midline incision provides adequate exposure for intracardiac defects. If more exposure is required, a sternotomy is performed after resecting the deformed cartilage, leaving the intercostal muscle attached to the sternum.[3,13]

Some authors have utilized a one-step surgery approach.[3,12-13,16] This can involve the minimally invasive Nuss procedure or the modified Ravitch procedure, which is still considered as superior by some authors, particularly when combine with open cardiac surgery through a midline sternotomy.[3,13-14,16] We decide to repair pectus in the same way as the Ravitch modification approach. The modifications are intended to improve access due to the existence of PE, which displaces the heart posteriorly and results in inadequate cardiac exposure. These modifications include the parasternal approach,[3,13] sternal turnover,[18] and median sternotomy.[16-17] Early wide cartilage dissection during the incision, before fixing the cardiac defect, improves the stability of the sternal retractor, provides optimal exposure for cardiac surgery, and facilitates the subsequent pectus repair procedure.[13,18] Excessive blood loss after costal cartilage resection and postcardiotomy coagulopathy must be managed carefully since they might delay sternal healing. In our example, the patient's first operation resulted in significant bleeding up to 1400 cc, compared to the more complex TOF operation, which only had 300 cc. Therefore, some surgeons recommend a stepwise approach to PE and congenital heart repair.[19] Some surgeons prefer a two-step procedure due to concerns about excessive bleeding, sternal infections, or lengthy surgical hours.17 However, if the surgery involves a complex cardiac procedure with high morbidity, a two-stage approach with the heart surgery is performed first is recommended.[3,13]

This strategy helps to minimize complications and allows for better management of the patient's condition throughout the surgical process, as we did it for our second patient. The use of temporary retrosternal bars in several studies has shown better outcomes in pectus repair by providing superior sternum stability, reducing pain and pulmonary complications, and yielding favorable long-term results.[3,13,18,20] This sternal fixation typically requires 6-12 months for the costal cartilage to develop in correct position.[19] Hasegawa et al. used Kirschner wires to provide firm fixation of the sternum, which has been shown to facilitate earlier union with superior primary osseous healing.[3] Additionally, preserving both internal mammary arteries can improve sternal viability and healing.[19] In the case of the first patient, we used plates and miniplates to maintain the stability of the sternum after removal of the cartilage. Fixation using convex Nuss Bars has also been reported by Okamura et al.[22] The Nuss procedure has significant downsides when combined with or delayed for cardiac surgery and pectus repair.[17,19] These include interference with cardiopulmonary resuscitation and planning for staged repair, potential sternal dehiscence, heart injury, pericardial effusions, and tissue adhesions. Additionally, it has been shown to reduce blood flow in internal mammary arteries postoperative, limiting their suitability for coronary bypass grafting.[17,19]

The ideal age for pectus surgery remains controversial. In our situation, both patients were teenagers and young adults. At this age, bone growth had already stopped, reducing the complication of earlier surgery at a young age. According to Fonkalsrud et al. the Ravitch procedure can be performed to correct pectus in children, as young as 3-6 years old. This timing is technically easier and takes less time than performing the surgery during adolescence and adulthood.[20] On the contrary, some authors believe that operating before the age of 4 or before growth completion increases the risk of recurrent sternal depression. If surgery is required, limit the amount of cartilage resected and avoid suturing the perichondral sheaths behind the sternum. Hysi et al. recommend doing two-step surgery on young patients, postponing the pectus correction until after the second growth spurt. However, in adults, they prefer concurrent surgery.[23] In summary, the timing and approach to surgical correction of pectus excavatum in patients with congenital heart disease must be individualized. Factors such as age, anatomical development, cardiopulmonary status, and the complexity of the cardiac defect should guide the decision between single-stage and two-stage procedures. While early intervention carries the risk of recurrence due to ongoing growth, delayed or staged surgery may offer better long-term stability and outcomes, particularly in complex or adult cases.

A limitation of this study is the small number of cases of patients with PE and CHD in our center. Therefore, we could only report two patients with both PE and CHD. Our study conducted using retrospective descriptive review. We treated patients according to the standard hospital service and the clinical presentation of the patient, so there was no intervention or control patients in our study.

CONCLUSIONS

The decision to perform a one-stage or two-stage procedure must be made carefully, as even patients with acyanotic CHD may present with complicating factors. For instance, in our first case, although the patient had an acyanotic defect, the individual presented in adulthood, by which time significant anatomical changes of the heart and pulmonary hypertension had already developed. These factors made the surgery more complex despite the acyanotic nature of the disease.

Therefore, we recommend that a two-stage procedure should still be considered for acyanotic patients who present with complicating factors, such as delayed diagnosis, structural cardiac changes, or pulmonary hypertension. In general, patients with complex cardiac conditions, regardless of cyanotic status, are better suited for a two-stage approach to reduce operative risks and improve outcomes.

CONFLICTS OF INTERESTS

The authors declare no conflicts of interest.

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