Aortic arch reconstruction using regional perfusion without circulatory arrest

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Abstract

Objectives: Deep hypothermic circulatory arrest during repair of aortic arch anomalies may induce neurological complications or myocardial injury. Regional cerebral and myocardial perfusion may eliminate those potential side effects. Methods: From March 2000 to March 2002, 48 neonates or infants with complex arch anomaly were operated on using the regional perfusion technique. Thirty-three patients were male and the median age was 24 days (range 5–301 days). Preoperative diagnosis consisted of coarctation or interruption of the aorta associated with ventricular septal defect (group I, n = 26) and arch anomaly with complex intracardiac defects such as hypoplastic left heart syndrome or its variants (group II, n = 22). Arterial cannula was inserted through the innominate artery and the flow rate was regulated to about 50–100 ml/kg per min during regional perfusion. Simultaneous myocardial perfusion was maintained using a Y-connected infusion line. Cardioplegia was applied during intracardiac repair. Results: Cardiopulmonary bypass and aortic cross-clamp times were 154 ± 49 and 39 ± 34 min, respectively. Temporary circulatory arrest for intracardiac procedures was performed in eight patients. However, the mean arrest time was minimized (range 1–18 min). The descending aorta clamping time was 33 ± 16 min. Operative mortality rates in each group were 0 and 18.2% (0/26 and 4/22). Late mortality rates were 0 and 11.1% (0/26 and 2/18) during 9.1 months of follow-up. Complications consisted of low cardiac output in eight cases, transient neurological problems in two cases, and transient renal insufficiency in two cases, respectively. Conclusions: Regional perfusion is feasible and can be used with acceptable results. It may reduce potential complications following aortic arch reconstruction using circulatory arrest. However, repair of aortic arch in the patients with complex intracardiac defects still imposes a significant rate of mortality and morbidity.

Keywords: Aortic arch repair; Regional perfusion

1. Introduction

Since the introduction of deep hypothermic total circulatory arrest (TCA) by Lewis and Tauffic [1] in 1953 and its application in the early complete repair of congenital heart anomalies by Barratt-Boyes et al. [2] in the 1970s, TCA has been widely accepted as an effective surgical adjunct in terms of decreased exposure to cardiopulmonary bypass (CPB), improved exposure, avoidance of multiple cannulas, and reduced edema [3]. However, TCA may induce neurological complications or myocardial injury [4–6]; therefore, many surgeons have attempted to improve the surgical result and to eliminate the neurological and developmental complications after complex repair of congenital heart anomalies using TCA. In these circumstances, continuous low flow bypass using regional perfusion (LFRP) was first introduced in 1996 by Asou et al. [7]. After then, McElhinney et al. [8], Pigula et al. [9], and Tchervenkov et al. [10] reported similar techniques using direct or indirect cannulation through the innominate artery. On the other hand, Sano and Mee [11] and Karl et al. [12] described an isolated myocardial perfusion technique to minimize the myocardial ischemia during TCA. Recently, Ishino et al. [13] reported a combined technique using LFRP and isolated myocardial perfusion in early primary repair of coarctation of aorta (CoA) and ventricular septal defect (VSD). However, optimal flow rate, temperature, and safe duration of LFRP are still questionable and the long-term results regarding neurodevelopmental outcome need to be investigated thoroughly.
The purpose of our study was to describe our surgical technique using LFRP and to investigate its effectiveness in view of cerebral and myocardial protection.

2. Materials and methods

2.1. Patients profile

From March 2000 to March 2002, 48 consecutive neonates or infants with complex arch anomaly were operated on using the regional perfusion and isolated myocardial perfusion technique. Thirty-three patients were male and the median age was 24 days (range 5–301 days). Body weight and body surface area were 3.5 ± 1.2 kg (range 1.6–8.3 kg) and 0.22 ± 0.05 m² (range 0.14–0.41 m²), respectively.

Preoperative diagnosis consisted of CoA or interruption of the aorta (IAA) associated with VSD (group I, Simple CoA, n = 26) and arch anomaly with complex intracardiac defects such as hypoplastic left heart syndrome (HLHS) or its variants (group II, Complex CoA, n = 22). The clinical characteristics and associated anomalies are summarized in Table 1.

2.2. Surgical technique

After the induction of general anesthesia and right radial arterial pressure monitoring, a standard midline sternotomy incision was made. The thymus was totally excised and the pericardium was incised and harvested for later use. Arch vessels, both pulmonary arteries, and patent ductus arteriosus (PDA) were carefully mobilized for cannulation and snaring. After full heparinization and purse-string sutures, 6Fr (2.0 mm) or 8Fr (2.7 mm) arterial cannula (RMI®, Edwards Lifesciences LLC, Irvine, CA) was inserted directly through the innominate artery and standard bicaval cannulation was instituted. In case of ductal dependent descending aortic circulation, 8Fr flexible arterial cannula (DLP®, Medtronic DLP, Grand Rapids, MI) was introduced at the proximal PDA and advanced into the descending aorta. These two arterial cannulas were Y-connected and CPB began. PDA was snared immediately after the bypass. A left ventricular vent (10Fr, DLP®, Medtronic, Inc., Minneapolis, MN) was introduced through the right upper pulmonary vein or left atrial appendage, if appropriate. The pH-stat strategy was used exclusively in acid-base management for cerebral protection. We took enough time to reach the desired core temperature. Usually, it took about 15–20 min [14]. During cooling, distal pulmonary arteries up to the second branch level, arch vessels, and descending aorta were extensively mobilized to relieve tension after anastomosis. Aortic root cannula (4Fr, DLP®, Medtronic Inc., Minneapolis, MN) was inserted and T-connected with the side hole of the innominate artery cannula.

When the rectal temperature reached 18 °C, the proximal innominate artery, left common carotid artery, and left subclavian artery were snared down to initiate the regional cerebral perfusion. Ascending aorta just distal to the root cannula was clamped for isolated myocardial perfusion. Descending aorta was also clamped as far distally as possible and gently elevated to produce a bloodless field and to reduce the anastomotic tension. The flow rate was regulated at about 50–100 ml/kg per min. The mean blood pressure of the right radial artery was maintained at about 40–50 mmHg and the mean hematocrit was maintained at around 20%. Simultaneous myocardial perfusion was maintained using a T-connected infusion line. Aortic arch repair was performed with extended end-to-side anastomosis of the descending aorta to the ascending aorta and aortic arch using a native

<table>
<thead>
<tr>
<th>Group</th>
<th>Total</th>
<th>Group I: simple CoA (n = 26)</th>
<th>Group II: complex CoA (n = 22)</th>
<th>P value</th>
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<tr>
<td>Demography</td>
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<tr>
<td>Sex (M/F)</td>
<td>33/15</td>
<td>19/7</td>
<td>14/8</td>
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<tr>
<td>Age (days)</td>
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<td>42 ± 55</td>
<td>46 ± 73</td>
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<tr>
<td>Body weight (kg)</td>
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<td>3.5 ± 1.3</td>
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<td>BSA (m²)</td>
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<td>Diagnosis (group I)</td>
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<tr>
<td>CoA with VSD</td>
<td>23</td>
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<tr>
<td>IAA with VSD</td>
<td>3</td>
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<td>Diagnosis (group II)</td>
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<tr>
<td>HLHS or variants</td>
<td>–</td>
<td>9</td>
<td>–</td>
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<tr>
<td>UVH</td>
<td>–</td>
<td>5</td>
<td>–</td>
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<tr>
<td>Taussig-Bing</td>
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<td>DORV</td>
<td>–</td>
<td>2</td>
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<tr>
<td>Posterior TGA</td>
<td>–</td>
<td>1</td>
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<tr>
<td>cc-TGA</td>
<td>–</td>
<td>1</td>
<td>–</td>
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<tr>
<td>Truncus arteriosus</td>
<td>–</td>
<td>1</td>
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a CoA, coarctation of aorta; BSA, body surface area; VSD, ventricular septal defect; IAA, interrupted aortic arch; HLHS, hypoplastic left heart syndrome; UVH, univentricular heart; DORV, double outlet right ventricle; TGA, transposition of the great arteries; cc-TGA, congenitally corrected transposition of the great arteries.
tissue-to-tissue technique, which was described by Fraser and Mee [15] and Rajasinghe et al. [16] (Fig. 1). In patients with HLHS, a triple anastomosis technique was used, which consisted of descending aorta, ascending aorta, aortic arch, and main pulmonary artery anastomosis [15].

After the aortic arch repair was completed, snares of arch vessels and descending aortic clamp were removed after complete de-airing, and the flow rate was fully restored (150–200 ml/kg per min). Myocardial perfusion was stopped and cardioplegia was applied for intracardiac repair.

2.3. Statistical analysis

Statistical analysis was performed with the Statistical Analysis System software package (version 6.12; SAS Institute, Cary, NC). The significance of differences between two groups was assessed by unpaired Student’s t-test, χ²-test or Fisher’s exact test. All results were expressed as the mean ± standard deviation and a value of P less than 0.05 was considered statistically significant.

3. Results

Operative and postoperative results are summarized in Table 2. CPB and aortic cross-clamping times were 154 ± 49 and 39 ± 34 min, respectively. Temporary circulatory arrest was applied in eight patients for the transfer of arterial cannula from the innominate artery to the reconstructed main pulmonary artery during the Norwood procedure (n = 3), closure of VSD in neonates with very small body weight and bilateral superior venae cavae (n = 2, 2.3 and 2.8 kg), closure of atrial septal defect (n = 1), evaluation of single atrioventricular valve (AVV) (n = 1), and atrial septectomy (n = 1). However, the mean arrest time was minimized (median 4.5 min, range 2–18 min). The descending aorta clamping time was 33 ± 16 min, which was significantly longer in the complex group (26 ± 7.6 min in group I versus 39 ± 20 min in group II, P = 0.01).

In group I, early primary repair was performed by arch repair and patch closure of VSD. In group II, associated procedures consisted of pulmonary arterial banding (PAB) with or without atrial septectomy in ten cases, modified Norwood procedure in six cases, one-stage biventricular repair with Norwood and Rastelli procedure in one case, Rastelli operation in one case, and aortic valvuloplasty with subaortic tissue excision in one case, respectively.

Operative mortality rates in each group were 0 and 18.2% (0/26 and 4/22, P = 0.02). The first patient was a 19-day-old female neonate weighing 3.2 kg who had unbalanced atrioventricular septal defect (AVSD) with severe arch hypoplasia. Due to the closure of PDA, metabolic acidosis and eventual cardiac arrest occurred preoperatively. She underwent emergent coarctoplasty, pulmonary arterial internal banding, and atrial septectomy. However, she suffered from acute renal failure, sepsis, and subsequent multiorgan failure postoperatively, and died. The second patient was a 12-day-old female neonate weighing 3.4 kg who had posterior transposition of the great arteries with severe arch hypoplasia. She also underwent palliative coarctoplasty with pulmonary arterial internal banding and atrial septectomy. Several days later, she underwent an arterial switch operation. She could not be weaned from CPB and we applied an extracorporeal membrane oxygenator (ECMO) for 16.3 h, but in vain. A coronary artery problem seemed to be the
cause of death. The third patient was a 10-day-old female neonate weighing 2.7 kg who had HLHS with mitral atresia, VSD, and obstructive total anomalous pulmonary venous return (TAPVR). She underwent a modified Norwood procedure with TAPVR repair. Postoperatively, she showed low cardiac output syndrome and eventually died after 3 h, maybe due to the pulmonary hypertensive crisis. The last patient was an 11-day-old male neonate weighing 3.2 kg who had HLHS with mitral and aortic atresia. He underwent a modified Norwood procedure and died on the first operative day because of low cardiac output syndrome.

Follow-up of all (44/48) operative survivors was completed for the mean duration of 9.1 ± 5.4 months (range 0.1–21 months). The mean duration of hospital stay was 28.8 ± 22.1 days (range 12–128 days). Late mortality rates were 0 and 11.1% (0/26 and 2/18) in each group (P = 0.08). One patient was a 10-month-old male infant weighing 6.5 kg who had functional single ventricle, criss-cross heart, subaortic stenosis, and type B interruption of the aortic arch. He underwent palliative aortic arch repair with conal septum resection, atrial septectomy, and pulmonary arterial internal banding. About 6 months after the first operation, he underwent bidirectional cavopulmonary shunt (BCPS) and died from pulmonary arterial thromboembolism about 3 months after the second operation. The other patient was a 29-day-old male neonate weighing 2.9 kg who had unbalanced AVSD, parachute left sided AVV with moderate regurgitation, and severe arch hypoplasia. He underwent palliative coarctoplasty with pulmonary arterial internal banding. About 7 months after the first operation, he underwent BCPS and subsequently received AVV repair and replacement due to progressive AVV regurgitation on the 27th and the 56th postoperative days, respectively. Eighteen days after the fourth operation, he expired due to low cardiac output syndrome.

Complications consisted of low cardiac output in eight cases, wound problems including one delayed sternal closure in five cases, sepsis in three cases, transient neurological problems in two cases, and transient renal insufficiency in two cases, respectively (Table 2). Among eight patients who showed postoperative low cardiac output syndrome, two had ECMO support. One in group I survived and the other in group II expired. Neurological complications occurred in two patients (4.2%). One infant with hemitruncus (right pulmonary artery from aorta) in group I showed transient choreoathetosis-like movement but completely recovered after 11 months of follow-up. The other patient with hypoplastic left heart complex and preoperative sepsis showed seizure-like movement but improved after 3 months of follow-up. Renal insufficiency occurred in two patients in group I, but the patients recovered shortly after peritoneal dialysis.

Recurrent aortic arch obstruction occurred in one patient. He was an 8-day-old neonate weighing 3.1 kg who had HLHS with aortic atresia, diminutive ascending aorta (diameter 1.5–2 mm), retrograde ascending aortic flow, severe arch and isthmic hypoplasia, large VSD, normal mitral valve, and normal sized left ventricle. He underwent one-stage biventricular repair that consisted of a modified Norwood procedure and Rastelli operation using homemade non-valved conduit. Five months after the operation, recoarctation developed and the pressure gradient was measured at about 74 mmHg. Successful balloon dilatation was performed and the pressure gradient dropped to 23 mmHg.

4. Discussion

Our study focused on the technical feasibility and effectiveness on postoperative outcomes after complex repair of
congenital heart defect with various arch anomalies using regional perfusion through innominate artery and coronary artery. The benefits of this technique have been well described by previous authors [7–10], particularly in terms of cerebral and myocardial protection during complex aortic arch repair. Ishino et al. [13] uniquely reported a combined technique similar to ours but their patients were mainly simple CoA with VSD. They used 30–50% (45–75 ml/kg per min) of full flow (150 ml/kg per min) and showed no neurological complication, no early mortality, and only one late death. Our study showed similar results without early or late mortality in the simple coarctation or interruption group. However, more complex anomalies such as HLHS or functional single ventricle with arch obstruction showed high morbidity or mortality rates.

Postoperative neurological complications after deep hypothermic TCA were well recognized and several authors proposed new techniques to avoid those potential side effects of TCA. Immediate postoperative and long-term neuro-developmental complications were systematically analyzed by the Boston Circulatory Arrest Group. However, only short-term results using the regional perfusion technique without circulatory arrest were reported. In our study, both early and late mortality rates were acceptable and significantly different in both simple and complex groups. Early postoperative neurological complications occurred in two patients (4.2%), which was also acceptable compared to the normally reported rates of 4–25% in TCA groups [17]. Fortunately, they recovered quickly during follow-up, but the long-term complications such as neuro-developmental abnormalities need to be evaluated further. In our opinion, the rate of neurologic complication was not so high. The number was only two, without significant statistical meaning. Individual characteristics such as associated anomalies in the central nervous system, poorly developed circle of Willis or other collaterals, or thromboembolic events seemed to be responsible for those neurological problems. Some reports showed no neurologic complication at all, but we could not fully understand those remarkable results. They could have occurred by chance. Our two patients had no abnormality on CT scanning of the brain and electroencephalogram. And they had fully recovered during the follow-up period, as described above. Actually we cannot imagine the cause of neurologic deficits, whether it be the improper technique (i.e. hyperperfusion) or any other factors such as preoperative hypoxic insult or poor collaterals.

Besides the use of regional perfusion, other factors such as pH-stat strategy for acid-base management, deep hypothermia, improved surgical skills, and comprehensive perioperative intensive care contributed to our improved short-term results. Recently, Jonas [3] emphasized that deep hypothermic circulatory arrest would be used more safely than innovative but unproven methods of regional perfusion unless clear guidelines regarding flow rate, temperature, and duration were established.

The optimal range of cerebral blood flow in deep hypothermic milieu (18 °C) is still debatable. Initially, Asou et al. [7] and McElhinney et al. [8] used flow rates of 50 and 30 ml/kg per min, respectively. Pigula et al. [18] objectively quantified that 20 ml/kg per min is sufficient using near-infrared spectroscopic analysis (NIRS). In a recent review, Pigula [19] has liberalized flow rates up to 40 ml/kg per min for both cerebral and somatic circulatory support. The neonates usually had larger brain volume than that of adults, weighing one-seventh to one-tenth of the body weight. Moreover, they had more extensive collateral circulation to the opposite side of the body including somatic circulation. Additionally, we used simultaneous myocardial perfusion combined with cerebral perfusion. It needed a higher flow rate than isolated cerebral perfusion (an additional myocardial blood flow of about 15 ml/kg per min; Sano and Mee [11]). Ishino et al. [13] used a flow rate of 30–50% of full flow and maintained arterial pressure at 30–45 mmHg using combined cerebral and myocardial perfusion in moderate hypothermia (28 °C). We used a somewhat higher flow rate of 50–100 ml/kg per min, which was similar to that of Ishino’s report [13]. It is well known that the cerebral blood flow is not related to rate, but related to pressure [20]. Therefore, the optimal flow rate during our technique might be much higher than that suggested above, in order to maintain perfusion pressure at about 40–50 mmHg. We considered that both cerebral and myocardial perfusion were effectively maintained in this higher flow rate. The risk of cerebral edema could be reduced using careful monitoring of right radial artery pressure and modified ultrafiltration. In our opinion, the higher the flow rate is, the better the clinical outcomes will be in terms of recovery of vital organ function as long as cerebral hyperperfusion does not occur. There is no evidence of definite cerebral hyperperfusion syndrome in our study populations.

Pigula et al. [21] reported that the somatic perfusion through various collaterals was maintained during LFRP with a flow rate of 30–40 ml/kg per min and suggested that the support of the subdiaphragmatic viscera should improve the ability of neonates to survive the postoperative period. Because of relatively rich collaterals and intact circle of Willis in the neonates, perfusion to the subdiaphragmatic viscera and left side of the brain was sufficient with such a low flow rate even if the mean perfusion pressure of the umbilical artery was maintained at 12 mmHg. In our study, the duration of regional perfusion was 33 ± 16 min, which was shorter than their report. However, the mean perfusion pressure of the right radial artery was maintained at about 40–50 mmHg, which was rather higher than theirs. We did not uniformly use the left radial artery pressure monitoring line. Usually there is some difficulty in peripheral arterial cannulation in small neonates or infants, and we cannot choose the monitoring site liberally. However, in our experience, an abundance of collaterals made it possible to maintain the perfusion pressure over 30 mmHg in the
contralateral side. Renal insufficiency occurred in only two patients (4.2%) and, as described earlier, they recovered soon after the peritoneal dialysis. But the direct comparison of their results showing no renal complication was inappropriate because of the difference in the sample size and various preoperative conditions. We considered that such a high flow rate would be beneficial in the functional recovery of somatic organs as well as cerebral protection.

Recent studies suggested that the higher hematocrit level would be beneficial on neurologic outcomes after the TCA or low flow bypass [22]. But there was some debate on the optimal hematocrit level and there has been no gold standard until now. We still believed that the lower hematocrit facilitated the cerebral blood flow and oxygen delivery with its rheologic benefits.

In our study, the rectal temperature was maintained at about 18°C. We agreed that the minimal temperature during regional cerebral perfusion would not be too profound as deep hypothermic circulatory arrest. Therefore, we used core-cooling as low as 28°C, similar to the usual moderate hypothermic CPB. But we used a unique separate air-cooling system of the operating room in order to protect the myocardium during cardiopulmonary arrest. So we did not use any topical myocardial cooling such as ice-slushed cold saline in order to prevent any cold injury to the myocardium or phrenic nerves. As a result, a low room temperature (about 10°C) inevitably reduced the core temperature of the small neonate or infant because of the surface cooling effect. Moreover, a lower temperature could protect the spinal cord during cross-clamping of the descending thoracic aorta. We believe that the deep hypothermia had beneficial effects on both myocardial and spinal protection, in addition to cerebral protection. We used both rectal and nasopharyngeal temperature only. Tympanic temperature cannot predict the brain temperature accurately [23]. It also has unacceptably low sensitivity, specificity, positive predictive value and negative predictive value in neonates and infants [24].

We exclusively used a direct cannulation method through the innominate artery even in the small neonates instead of an indirect method using synthetic graft made of polytetrafluoroethylene. We considered it a very simple and reproducible method which did not obscure the surgical field. In addition, there was no cannulation-related complication such as stenosis or intimal injury. Usually, the size of the innominate artery was sufficient for cannulation and we easily inserted the 6Fr or 8Fr arterial cannula (RMI®, Edwards Lifesciences LLC, Irvine, CA). Even in an 18-day-old full-term neonate weighing 1.6 kg who had a functional single ventricle with subaortic obstruction, we introduced a 16-gauze angiographic catheter into the innominate artery without any problem. He received a modified Damus–Kaye–Stansel procedure 6 months after the first operation and subsequently received BCPS 14 months later. He is now waiting for a Fontan operation and angiography shows a widely patent innominate artery. For neonates with HLHS undergoing stage I Norwood palliation, we moved the cannula to the neo-aorta and constructed the shunt at the more distal innominate artery. We believe that any injury to the inner surface of a small polytetrafluoroethylene graft can compromise the shunt function, especially thrombotic occlusion. Therefore, we did not cannulate the shunt directly.

We used separate myocardial perfusion during aortic arch reconstruction. In our deep hypothermic temperature setting, the heart maintained extremely slow and empty beating, showing no signs of dilatation or functional derangement. Actually we had no comparative data between myocardial perfusion and cardioplegic arrest. But we believed that our myocardial perfusion technique practically reduced the myocardial ischemic time and gave us more time during intracardiac repair.

The limitations of our study were the usual problems in retrospective studies, lack of precise neurological examination specific to the neonates or infants, and non-randomized uncontrolled patient groups. Because the time span using TCA is quite different from that of regional perfusion, it has little meaning in comparing the neurologic outcome between two techniques. To evaluate the long-term effectiveness of our technique on the neuro-developmental outcome, a prospectively randomized controlled study with precise neurological examination and perioperative monitoring is necessary.

In conclusion, regional perfusion is feasible and can be used with acceptable results, which are comparable to those reported from many institutions using circulatory arrest. It may reduce the potential complications following aortic arch reconstruction using circulatory arrest. However, repair of the aortic arch in patients with complex intracardiac defects still has a significant rate of mortality and morbidity.

Acknowledgements

This study was approved by the Institutional Review Board of our hospital.

References


