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Catastrophic events associated to the surgical treatment of ostium secundum atrial septal defects. Reasons for not underestimating this type of congenital cardiopathy

Eventos catastróficos associados ao tratamento da comunicação interatrial tipo ostium secundum: razões para não se subestimar este tipo de cardiopatia congênita

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Abstract

***Objective*: The present article was motivated by the frustrating experiences with four patients who underwent surgical treatment of ostium secundum atrial septal defect (ASD-II) and who died in extremely dramatic circumstances.**

***Method*: This is a retrospective study based on clinical data. The bibliographical research included: paradoxical thromboembolism (cerebral, pulmonary or mesenteric), central nervous system vascular malformations and anomalous vena cava connections. This research was based on possible events, unexpected and catastrophic, that could have directly caused the patients' deaths.**

***Results:* All patients were female. The operations were performed under cardiopulmonary bypasses with less than 20 minutes of aortic cross-clamping. The causes of death were: non-occlusive intestinal ischemia, rupture of a cerebral aneurysm of the anterior communicating artery, cor pulmonale associated with pulmonary arterial hypertension and thromboembolism and probable cerebral thromboembolism in a child that needed to be reoperated to correct anomalous inferior vena cava drainage into the left atrium.**

***Conclusion*: Even being technically easy to treat, death due to atrial septal defects in extremely dramatic circumstances is possible, so "Do not underestimate atrial septal defects in congenital heart surgery".**

***Descriptors:* Heart septal defects, atrial, surgery, complications. Heart defects, congenital, surgery, complications. Extracorporeal circulation.**

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Resumo

Objetivo: **O presente trabalho clínico foi motivado pela frustrante experiência de quatro pacientes operados para o tratamento cirúrgico da comunicação interatrial tipo ostium secundum (CIA-II), que vieram a falecer em condições extremamente dramáticas.**

Método: **Estudo retrospectivo embasado em dados de prontuários. As pesquisas bibliográficas incluíram: tromboembolismo paradoxal (cerebral, pulmonar ou mesentérico), malformações vasculares do sistema nervoso central e conexões anômalas das veias cavas. Estas pesquisas da literatura foram embasadas em possíveis eventos, inesperados e catastróficos, que levaram quatro pacientes ao óbito.**

Resultados: **Os quatros pacientes, todos do sexo feminino, foram submetidos a atriosseptorrafia com tempo de parada cardíaca isquêmica inferior a 20 minutos, em circulação extracorpórea. As causas de óbito foram: isquemia intestinal não-oclusiva, ruptura de aneurisma cerebral de artéria comunicante anterior, cor pulmonale com hipertensão arterial e tromboembolismo e um provável tromboembolismo cerebral em uma criança que precisou ser reoperada pela drenagem da veia cava inferior em átrio esquerdo.**

Conclusão: **A lição final deste trabalho é: "Não subestime a comunicação interatrial em cirurgia cardíaca!".**

Descritores: **Defeitos do septo interatrial, cirurgia, complicações. Cardiopatias congênitas, cirurgia, complicações. Circulação extracorpórea.**

INTRODUCTION

Ostium secundum atrial septal defect (ASD-II) is one of the most frequent congenital cardiopathies. Although compatible with an uneventful evolution up to the sixth decade of life, it is very well established that patients who have ASD-II have higher morbidity rates and shorter life expectancy compared with the normal population. For this reason, its closure is recommended in the preschool age, even if asymptomatic, or soon after it is diagnosed [1-3].

The present article was motivated by the frustrating experiences with four patients operated on for closure of the ASD-II, who died in extremely dramatic circumstances. All patients were female. The operations were performed under cardiopulmonary bypasses with less than 20 minutes of aortic cross-clamping time. The article of GUNAY [4] summarizes in its title the whole idea of this text: "Do not underestimate the atrial septal defects in congenital heart surgery".

CASE REPORTS

Case 1. A 22-year-old white woman presenting palpitations and dyspnea to habitual efforts over a period of four years was referred for surgical treatment. During her first pregnancy the symptoms increased. She presented with an audible systolic murmur at a pulmonary focus, and an incomplete right bundle branch block on the ECG.

In May 1982, after cesarean delivery, she was submitted to heart catheterization that revealed the presence of an ASD-II with left to right shunt of 2.5/1. However, the operation had to be postponed because of a new pregnancy just after the breast-feeding period. A c-section was performed at the term of this pregnancy.

The ASD was closed by direct running sutures using crystalloid cardioplegia for myocardial protection. The total time of surgery was 80 minutes and the cardiopulmonary bypass (CPB) time was 30 minutes with 15 minutes of aortic cross-clamping time.

On the third day, after discharge from the intensive care unit, the patient presented sudden arterial hypotension, tachycardia and signs of intense peripheral vasodilatation. The patient was treated with volumetric expansion and amines, becoming hemodynamically stable after two days. Soon after, the patient complained of abdominal pain and diarrhea with dark and fetid feces. A blood analysis showed pronounced leukocytosis, increased levels of bilirubin and transaminases (SGTP and SGOT). On the eighth postoperative day, the fifth day of persistent abdominal pain,

the patient became dyspneic. A laparotomy revealed massive intestinal necrosis. A broad intestinal resection was performed, leaving about 60 cm of the small bowels, part of the descending colon and the sigmoid (Figure 1). The patient was maintained under parenteral nutrition and died on the sixth postoperative day of multiple organ failure caused by septicemia. The necropsy revealed absence of thrombosis in non-inflamed areas, associated with areas of intense necrosis but without any anatomical pattern, as well as lesions focused on the mucous membrane, strongly suggestive of non-occlusive intestinal ischemia.

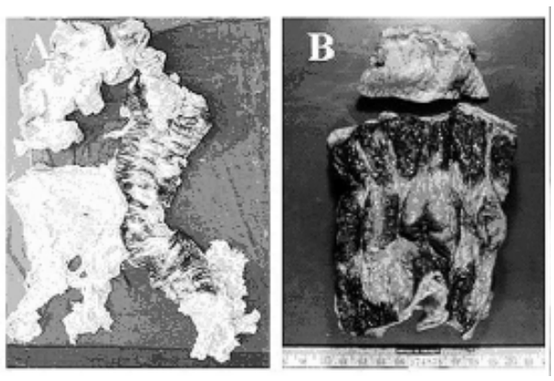

Fig. 1 - Case 1: A - Resected intestinal bowels; B Probable melanosis coli pigmentation

Case 2: A 31-year-old white woman with an ASD II was admitted to our institution for surgical treatment. Her main symptoms were dyspnea and palpitation to medium and great efforts. The physical examination revealed slight systolic murmur at a pulmonary focus and the ECG an incomplete right bundle branch block. The chest radiography showed increased pulmonary vascularity. An ASD-II was clearly identified by Echo Doppler and heart catheterization. In March 1989 the ASD-II (4 cm x 2 cm) was surgically closed under CPB without any surgical incident. The surgery lasted 135 minutes, the CPB time was 35 minutes and the aortic cross-clamped time was 20 minutes. About two hours after surgery, the patient presented a hypertensive crisis, with mean arterial pressure over 200 mmHg, followed by a brief convulsion, accentuated bradycardia, fixed mydriasis and areflexia. The liquor examination suggested intracranial hemorrhage. A carotid angiogram revealed the rupture of an extensive aneurysm of the anterior communicating artery (Figure 2). No therapeutic measure had effect and the patient remained in deep coma, areflexia, paralytic mydriasis, evolving to death on the third postoperative day due to electromechanical heart dissociation.

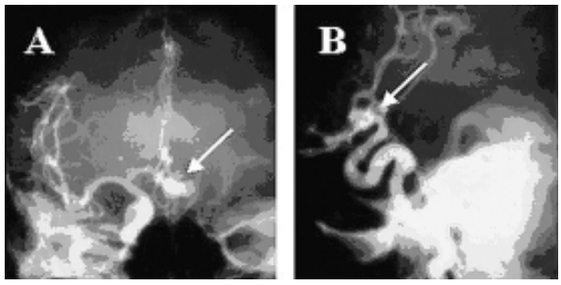

Fig. 2 - Case 2: A – carotid angiogram (posterior-anterior perspective) and; B - Carotid angiogram (lateral perspective). The arrows denote the aneurysm of the anterior communicating artery.

Case 3. A 6-year-old child who was receiving medical attention since she was about 3 years old for frequent episodes of bronchospasm, whose frequency had been increasing, was referred to our institution to have an ASD-II surgically closed. The diagnosis had been made by echocardiography.

She underwent an operation in August 1992. During the operation an ASD in a higher position than normally is expected for ASD-IIs was observed. The right atrium was distended and cannulation of the inferior vena cava was difficult. All the pulmonary veins were connected to the left atrium, with the upper veins in a higher position and the lower veins connected more to the left. The closure was performed using a double running suture with polypropylene 4-0. During CPB poor venous drainage to the oxygenator was evident, and the anesthesiologist noticed edema and cyanosis of the child's face. The surgery was completed as fast as possible, with aortic cross-clamping time of 15 minutes and CPB time of 20 minutes. She recovered from CPB with spontaneous sinus rhythm and good hemodynamic conditions. Her left atrium pressure was normal, but she was cyanosed and with bronchospasm. In the intensive care unit her central venous pressure became extremely high, consequently a new catheter was positioned in the inferior vena cava revealing venous pressure of 10 cm H_2O. In spite of the suspicion of an obstruction of the superior vena cava, surgical intervention was postponed, since the child was hemodynamically stable. On the tenth postoperative day, still with cyanosis more noticeable on the trunk and upper limbs, the child suffered a stroke, with right hemiplegia, which improved with physiotherapy.

A heart catheterization revealed a slight stenosis of the superior vena cava, absence of any intracardiac shunt and drainage of the inferior vena cava (IVC) to the left atrium (Figure 3). A second operation was scheduled. Through a right atriotomy the ASD was opened and a bovine pericardium patch was sewed allowing the re-direction of the IVC flow to the right atrium, thereby reconnecting the

inferior vena cava to the right atrium. The immediate postoperative evolution was uneventful until the fifth postoperative day when the child presented asystole unresponsive to all maneuvers of advanced cardiac life support. The family did not allow a necropsy. The probable cause of death was thromboembolism to the central nervous system (most probable) or pulmonary thromboembolism.

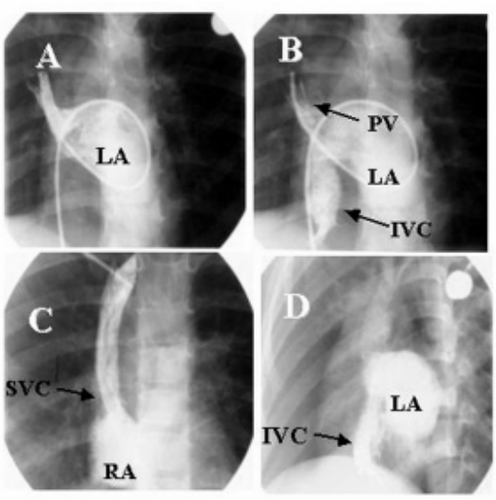

Fig. 3 - Case 3: A and B-Catheter introduced by the femoral vein until the right superior pulmonary vein (PV) contrasting in this phase the left atrium (LA). The angiography demonstrates the inferior vena cava drainage in the left atrium (LA); C-contrast injection in the superior vena cava (SVC) with normal drainage in the right atrium (RA); D-Left anterior oblique incidence showing the inferior vena cava (IVC) drainage in the left atrium (LA).

Case 4: A 26-year-old woman with clinical history of at least one year of fatigue to great and medium efforts was referred to our institution. An ECG showed signs of right ventricle overload, the chest radiogram showed increased pulmonary vascularity and the echo Doppler evidenced an ASD-II with estimated pulmonary arterial pressure of 90-100 mmHg.

In August 1993 the ASD-II was closed by direct double running suture without using cardioplegic solution. The time of surgery was two hours, the CPB time was 30 minutes and aortic cross-clamping time was 15 minutes. The postoperative evolution was uneventful and the patient was discharged four days later.

About a year later, after an uneventful pregnancy, she started to complain of mild dyspnea, however with no signs of heart failure and the oxygen saturation, measured by pulse oximetry, was always greater than 94%. A Doppler echocardiogram, four years after the surgery did not reveal any residual defect and her mean pulmonary arterial pressure had reduced to 53 mmHg. In March 1999 she was asymptomatic, but in December of the same year she returned with dyspnea to great efforts with progressive deterioration. She was acyanotic, but the chest auscultation revealed a hyper-phonetic second heart sound (S2) at a pulmonary focus and the arterial blood saturation, measured by pulse oximetry, was 89% with reductions to 86% when submitted to isometric effort. A chest radiogram revealed increased right cardiac chambers, dilated pulmonary artery trunk and poor peripheral lung circulation. The ECG showed great overload of the right cavities with "pulmonale" P wave. Echocardiography revealed severe "cor pulmonale". The lung spirometry was normal, but the arterial oxygen saturation was 90% and the partial pressure of oxygen was 62 mmHg. A ventilation/perfusion lung scintigraphy revealed a high probability of pulmonary thromboembolism. Computerized tomography and magnetic resonance imaging (MRI) (Figure 4) also suggested pulmonary emboli. Further investigations were negative for lupus anticoagulant factor, C-protein activity, antithrombin III, total and free S-protein

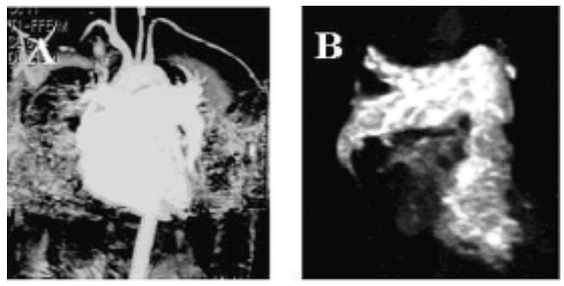

Fig. 4 - Case 4: Magnetic resonance imaging (MRI) – A – Pulmonary artery diameter two times bigger than aorta diameter; B- Pulmonary artery left branch with suggestive images of organized thrombi.

(antigen) and anticardiolipin antibodies.

The following treatments were considered: a) Clinical treatment with pulmonary vasodilators (nifedipine, ACE inhibitor, quinapril) and domiciliary oxygen therapy in the near future; b) Cardiopulmonary transplant; c) Bilateral lung transplantation; d) Unilateral lung transplantation and e) Pulmonary thromboendarterectomy. We opted for thromboendarterectomy.

The surgery was carried out in February 2000, using CPB, hypothermia of 23 ºC and low flow with short periods of circulatory arrest, according the University of San Diego protocol. The operation revealed much enlargement of the

right heart chambers and pulmonary artery dilation (about twice the size of the aorta). Unexpectedly a small ASD was discovered (about 1.5 x 1.5 cm), close to the inferior vena cava. The ASD was sutured, and soon as the pulmonary artery was opened the possibility of improvement of the pulmonary hypertension with right heart relief was questioned. The thrombi were not compatible with the images obtained by the chest tomography and MRI. The thromboendarterectomy was quite convincing including some tertiary branches of the left pulmonary artery. There were no thrombi in the right branch, which was dilated and atherosclerotic. The surgery was carried out without aorta cross-clamping and the CPB time was 220 minutes.

The postoperative course was catastrophic. The patient developed pulmonary hypertension of up to 180 mmHg, which was not responsive to oxygen using a respirator and the use of vasodilators, either by vein or inhalation, was totally ineffective. On the second day the situation was unsustainable and a surgical septectomy, using venous stasis, was carried out. The operation took a very short time, but the patient became extremely cyanotic and a few minutes later she presented irreversible electromechanical dissociation.

DISCUSSION

Case 1. ASD and digestive complications after heart surgery

Abdominal complications related to cardiopulmonary bypasses are relatively rare and blamed for less than 1% of all of complications which has been decreasing over the last decades [5]. On the other hand, this incidence increases up to six times depending on the complexity of the heart operation [6]. The most frequent gastrointestinal complications are: acute esophagitis, digestive tract hemorrhages, postoperative jaundice, acute inflammatory abdomen and intestinal obstruction. Ischemia and intestinal infarction are less frequent and usually associated with aortic insufficiency, atherosclerotic disease and ages between 40 and 50 years [7]. The present case is unique in the literature, associating mesenteric ischemia with an operation to close an ASD.

The possible causes of intestinal ischemia and necrosis in this case are a matter of speculation. Usually, it is very difficult to point the primary cause of any occurrence in a surgery of short duration. On the third postoperative day the patient presented a sudden episode of hypotension, palpitations and signs of peripheral vasodilatation. The doubt is if the subsequent events were the cause or consequence of the intestinal ischemia. These doubts are common to all similar cases described in the literature. The majority of reported cases were submitted to laparotomy with extensive intestinal resections. The importance of ceasing use of digitalis and vasoconstrictors and the use of direct or epidural splanchnic blockades associated to the use of á-blockers (such as fenoxybenzamine) has been emphasized. In spite of the kind of treatment the mortality rate is close to 100%, partially due to the delay in diagnosis, since in the majority of the patients intestinal murmurs and movement are present. Abdominal pain is the main symptom. This situation, late diagnosis and sepsis happened in this patient.

The reason that the incidence of non-occlusive mesenteric infarction is low in patients that present very low cardiac output after operations involving CPB remains unknown, especially among the patients that use intra-aortic balloon pumps and high doses of catecholamines. These facts suggest that a particular physiopathologic mechanism and individual susceptibility are responsible for this catastrophic event.

The best therapy is still debatable. Although in the majority of the cases described in the literature a laparotomy was performed, there are two reports of successful conservative treatment using local vasodilatation [8,9].

Case 2. ASD and associated with Central Nervous System malformations

The number of patients who have congenital heart malformations who enter adolescence and become adults has been increasing. A Mayo Clinic experiment concluded that patients with congenital heart malformations have a higher risk of suffering intracerebral aneurysms and arterial dissections, particularly during adolescence. The intracranial and cervical muscle arteries are derived from neural crest cells, which also are important in the early development of the heart. The existence of this common embryological structure may be relevant to the association between congenital heart diseases and arterial malformations of the cephalic segment [10]. It is important to notice that the Mayo Clinic series, highly regarded by the quality of its documentation, did not mention any cases of ASD among the 14 patients with congenital heart malformations. Our reported case is unique among the 7000 heart operations we have performed, revealing the rarity of this association. A wide-ranging review of publications revealed only one similar case from India in which the association of ASD and intracerebral aneurysm was reported [11].

In the reported case the diagnosis was made by a clinical neurological examination, analyses of the liquor and by a carotid angiography. At the time of the event we did not have access to more sophisticated and less invasive diagnostic procedures. Since the patient presented signs of cerebral death, no further investigations were carried out.

Case 3. ASD and inferior vena cava drainage to the left atrium

The anomalous drainage of the inferior vena cava (IVC) to the left atrium is rare. A bibliographical investigation of the database MEDLINE revealed fewer than 50 cases since 1966. This anomaly is due to the failure of the embryonic development of the right sinus venous valve, septum secundum and the dorsal interatrial septum portion [12].

It is important to emphasize the possibility of surgical closure of a prominent Eustachian valve mistakenly considered part of the atrial septum [13, 14]. Additionally, the IVC anomalous drainage to the left atrium may be associated to other congenital cardiopathies and not just an ASD. Although this is rare, associations have been described with the Tetralogy of Fallot [15] and ASD associated to anomalous drainage of the pulmonary veins [16].

Usually the preoperative diagnosis is made using angiocardiographic or Doppler echocardiographic studies, with sophistications such as echocardiographic contrast or three-dimensional images [17]. MRI is other diagnostic tool that may be used for the diagnosis of IVC anomalous drainage to the left atrium. Although, there is a consensus that an ASD-II may be operated on based only on the conventional Doppler echocardiographic diagnosis, medical literature reports few cases whose diagnoses of this systemic venous anomaly has been made preoperatively. Cyanosis associated with normal pulmonary pressure in newborn babies is indicative of this anomaly. In the largest series, mentioned above, the cardiologists' group, certainly considered this type of congenital malformation, since the cases were diagnosed preoperatively [18].

The present case has characteristics compatible with the data reviewed and presented as part of this discussion. The operation was indicated based on clinical signs, age and anatomical anomaly diagnosed by Doppler echocardiography. The fact that the cannulation of the IVC was possible when establishing the CPB circuit suggests the existence of a redundant Eustaquian valve or the possibility that a supra-hepatic vein drained independent of the anomalous IVC directly to the right atrium. It is still possible, that the IVC connection to the left atrium was iatrogenic. The persistence of cyanosis and a certain degree of facial edema led to the decision to perform heart catheterism that revealed the presence of the IVC connection to the left atrium. The child was reoperated, and the IVC drainage corrected using a bovine pericardium patch. After this surgery the cyanosis and the face edema disappeared, the child became hyperactive again, but, unfortunately she died suddenly, with electromechanical dissociation of the heart, probably due to pulmonary thromboembolism or, even more likely due to cerebral thromboembolism. Pulmonary thromboembolism as a cause of sudden death in the postoperative period of a child was recently demonstrated by an autopsy that revealed thrombotic deposits along the suture line of the ASD-II [19].

Case 4. CIA and thromboembolism

Cerebral Thromboembolism

Paradoxical thromboembolism may occur in any part of the body, but it is particularly serious when it affects the brain or lungs. The patent oval foramen (POF), an embryological reminiscent found in 27% of adults, is a potential site of right-to-left intracardiac shunts. This shunt can be the result of a reversal of the pressure gradient or abnormal blood inflow to the right atrium. The pathological consequences of the right-to-left shunt include hypoxemia and paradoxical embolism. Paradoxical embolism through a POF is well documented and its role in the etiology of thromboembolism of obscure origin (cryptogenic thromboembolism) is a matter of great discussions and controversies [20].

Pulmonary Thromboembolism

Pulmonary embolism, although less frequent than systemic embolism, may also be a catastrophic occurrence associated with ASD, even after corrective surgery. Helicoidal computed tomography and MRI, as well as pulmonary arteriography, are excellent methods of diagnosis by imaging.

Also, an isolated pulmonary thromboembolism event is possible in patients with ASD. This possibility has been reported more frequently in elderly patients. There is no precise data about the prevalence of pulmonary hypertension caused by thromboembolism. In the University of San Diego experience with the largest series of patients undergoing pulmonary thromboendarterectomy in the world, the occurrence of ASD or POF was around 25% of all operated cases [21].

The surgical options are pulmonary thromboendarterectomy and lung or cardiopulmonary transplantation. There are reports of accentuated improvement in right heart function following unilateral lung transplantation.

The patient reported here had her ASD corrected by direct running sutures about six years before. She always presented physical discomfort incompatible with her physical activity. This symptom was only considered when the patient presented 88% of oxygen saturation measured by pulse oximetry. This unexpected data triggered an investigation, which revealed severe

pulmonary hypertension and right heart failure due to chronic pulmonary thromboembolism. The reasoning was all based on speculation. The only risk factor, besides the ASD, was the use of contraceptives. One fact became absolutely clear, there was an underestimation of the values of 90-100 mmHg, which were very high for an ASD of 2.2 cm, before the ASD surgical correction. Our hypotheses are: a) that this patient a victim of non-detected episodes of small pulmonary embolic episodes or pulmonary endothelium dysfunction. But 2-3 years after the surgery the arterial pulmonary pressure, measured by echo Doppler, was around 50 mmHg and an investigation of congenital defects of the coagulation was negative; b) that this patient a victim of pulmonary thromboembolism from the interatrial septum sutures. This hypothesis is unlikely, as the immediate postoperative period was uneventful and; c) that new thromboembolic episodes occurred that became more frequent or larger after ASD closure, as at that time the pulmonary arterial pressure was around 50 mmHg, against 90-100 mmHg in the preoperative period and 103 mmHg at the final event. It is important to remember that a small ASD was found, which did not seem to be residual, located very close of the IVC entrance to the right atrium. This residual defect would be a source of paradoxical pulmonary thromboembolism.

The main motivation of this text is implicit in its title. Lethal complications of ASD and its surgical correction are rare. Are any factors important for this kind of observation? Maybe cases are rarely reported and contribute to the paradigm that surgical treatment of ASD is "a task for residents?" In any case this is a warning as previously mentioned: "do not underestimate the simplest congenital pathologies in heart surgery." The four cases reported here are strong examples of this assertion. The catastrophic lethal events were: non-occlusive intestinal ischemia, rupture of a cerebral aneurysm of the anterior communicating artery, cor pulmonale associated with arterial pulmonary hypertension and thromboembolism and probable cerebral thromboembolism in a child that needed to be reoperated to correct anomalous inferior vena cava drainage into the left atrium.

A mandatory question concerns to the prevention of such fatal events. The answer is difficult, because the methodology of surgical treatment of ASD with cardiopulmonary bypass is very well established. If there is no negligence to the details, complications as presented would only be due to tragic and unusual fatalities.

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