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Case Report

Cardiocentro ERNESTO CHE GUEVARA SANTA CLARA CUBA

False aneurysm of ascending aortic cannulation site after cardiac surgery. Case report and literature review

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Abbreviations
CPB: cardiopulmonary bypass
TTE: transthoracic echocardiogram
FAA: false aortic aneurysm
SICU: Surgical Intensive Care Unit

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ABSTRACT

False ascending aortic aneurysm, as a complication in cardiac surgery, has an incidence of less than 0.5%, but a mortality that ranges from 6.7% to 60%. Aortic manipulation and infection are the main predisposing factors. Its clinical presentation is non-specific and may be found incidentally on subsequent check-ups. The case of a patient undergoing mitral valve surgery who was diagnosed with ascending aortic false aneurysm, eleven months after postoperative period, requiring emergency surgical treatment is presented. The surgery was performed using extra-mediastinal peripheral cannulation, without induced circulatory arrest or hypothermia for proper dissection. The case shows a technical version to face an uncommon but highly challenging disease as it needs re-interventions and successful surgical strategies.

Keywords: False aneurysm, Ascending aorta, Postoperative complications, Cardiovascular surgery, Thoracic surgery, Induced hypothermia

Falso aneurisma del sitio de canulación aórtico después de cirugía cardíaca: Informe de un caso y revisión de la literatura

RESUMEN

El pseudoaneurisma de aorta ascendente, como complicación en cirugía cardíaca, tiene una incidencia menor del 0,5%, pero una mortalidad que varía del 6,7 al 60%. La manipulación aórtica y la infección son los principales factores predisponentes. Su clínica puede ser variada y constituir un hallazgo casual en controles posteriores. Se presenta el caso de una paciente operada de cirugía valvular mitral que a los 11 meses del postoperatorio se le diagnostica un pseudoaneurisma de aorta ascendente, que requirió tratamiento quirúrgico de emergencia. La cirugía fue llevada a cabo con la utilización de canulación periférica extramediastínica, sin requerir parada circulatoria, ni hipotermia para su correcta disección. El caso muestra una variante técnica para enfrentar una enfermedad poco frecuente, pero altamente desafiante, por la necesidad de reintervención y una correcta estrategia quirúrgica.

Palabras clave: Falso aneurisma, Aorta ascendente, Complicaciones postoperatorias, Cirugía cardiovascular, Cirugía torácica, Hipotermia inducida

INTRODUCTION

Pseudoaneurysm or false aortic aneurysm (FAA) is a serious complication

that may occur several months or years after heart surgery[1]. It is defined as a blood-containing injury resulting from arterial wall interruption with blood extravasation which is contained by periarterial connective tissue[2]. It is caused by the disruption of at least one layer of the vessel wall at the cannulation site or along the aortic suture lines. Initially, the FAA is surrounded and controlled by the remaining vascular layers or closely related structures of the mediastinum with or without development of a neointima.

When the pressure of the aortic false aneurysm exceeds the maximally tolerated wall tension of the surrounding tissue, fatal rupture occurs. Progressive widening of the false aneurysm may cause other feared complications such as a bleeding fistula and compression or erosion of neighboring structures. Aortic dissection, infections, degenerative arterial wall, trauma, excessive use of biological glue and a poor surgical technique are recognized as risk factors[2-4]. Surgical treatment remains the solution, despite rapid advances in endovascular techniques.

A technical version without the use of induced hypothermia or cardiac arrest performed by our surgical team to manage this dangerous complication in a patient 11 months after heart valve surgery is described herein.

CASE REPORT

We present the case of a 51-year-old, mixed race, obese, ex-smoker woman with a history of hypertension and rheumatic fever in childhood with severe mitral valve involvement (double mitral injury with predominance of insufficiency) who was referred to our Center for surgical treatment. Mitral valve replacement was performed with a 27 mm bileaflet mechanical prosthesis (St. Jude Medical Inc., MN, USA), after longitudinal median sternotomy, with ascending aortic and vena cava cannulation (according to hospital management protocols) requiring 72-minute aortic clamping and 105-minute cardiopulmonary bypass (CPB). The patient had a slow, eventful clinical course in the Intensive Care Unit (ICU) with hypoxemia, prolonged mechanical ventilation and ventilator-associated pneumonia (Klebsiella pneumoniae) which resolved after recommended antimicrobial treatment. Several surgical procedures were necessary for drainage of pleural effusions and she was discharged two months post operatively with standard treatment.

Outpatient follow-up showed little control of oral anticoagulation (warfarin) and the target INR (International Normalized Ratio) range of 2.5-3.5 was difficult to achieve. Follow-up transthoracic echocardiogram (TTE) showed no abnormalities in prosthetic functioning, nor paracardiac masses or thrombus; biventricular function was normal; however, eleven months after surgery she was re-admitted to a nearby municipal hospital; she was febrile and complained of dyspnea, dysphagia, and oppressive chest pain. On physical examination she presented a fairly audible prosthetic click, without heart murmurs, bibasal wet rales, hematuria, extensive-chest, abdomen or limbs- ecchymosis and an INR>10. She was referred to the provincial hospital for alleged risk of bleeding related to the dicumarinics. She worsened clinically with diagnosis of severe respiratory sepsis; hence, she underwent antimicrobial treatment and was promptly transferred to our center.

She was admitted to the SICU with hemodynamic deterioration and mechanical ventilation due to acute respiratory failure. Chest x-ray showed significant mediastinal enlargement and the electrocardiogram revealed atrial fibrillation with rapid ventricular response. The TTE demonstrated: normo-functioning prosthesis, without pathological gradients, leaks, thrombus or vegetations; with a large, echolucent, sharply-outlined image that made us suspect the presence of a massive FAA. Urgent multislice contrast computed tomography enabled us to define the diagnosis and action strategy according to the characteristics of the complication (**Fig. 1**).

Hemodynamic deterioration and the imminent risk of rupture of the FAA prompted urgent surgery. The presence of a relatively small entrance hole in the ascending aorta and the experience of our working group led to the strategy of trying to assess the FAA without temperature drop, or total circulatory arrest. Being close to the sternum, they chose to use ECC CPB with femoro-femoral peripheral cannulation prior to re-sternotomy (**Fig. 2**). A venous cannula was placed in the right atrium via left femoral vein, and another in the left common femoral artery for arterial perfusion, which allowed lowering the FAA internal pressure and reducing the risks of reopening accidents.

Once the cardiopulmonary bypass was implanted, sternal reopening with an oscillating saw and careful dissection of tissues surrounding the false aneurysm were performed, until it was completely exposed for opening and locating the aortic rupture

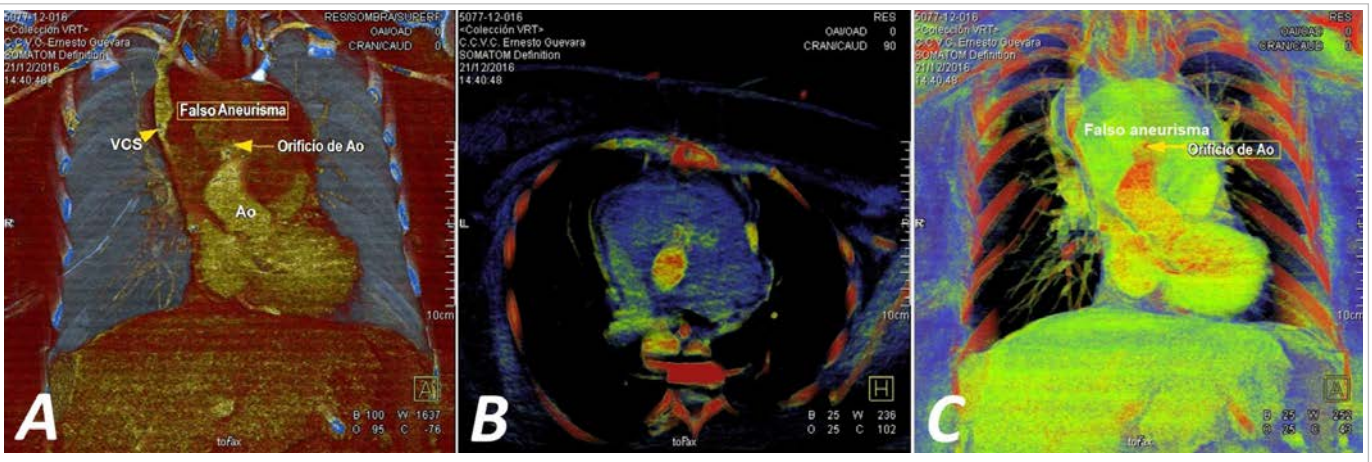

Fig. 1. Tomographic images evidencing interrupted aortic-wall continuity where the hematoma is enclosed by the neighboring mediastinal organs. **A.** Anteroposterior view. Note compression of the superior vena cava (VCS). **B.** Axial cut. **C.** Anteroposterior view. Ao, aorta.

site, which was found in the cannulation area for arterial perfusion. The lead surgeon proceeded to gently occlude it with his index finger allowing a surgical "cul-de-sac" around it. Once controlled, the perfusion pressure was lowered (mean pressure not less than 50 mmHg) to knot and place Prolene® 3/0 reinforcements with Teflon strips (**Fig. 3**).

Perfusion pressure was restored to the usual levels for normothermal surgery, the sutures corresponding to the other cannulation sites were inspected and no signs of sepsis were found. The FAA was partially resected, and samples were taken for culture. The extracorporeal circulation time was 93 min. The surgical procedure was routinely accomplished and the patient was transferred to the SICU. She improved hemodynamically and was successfully weaned off oxygen support after 72 hours. The suggested antibiotic treatment was completed and she was discharged 25 days after reintervention. Microbiological studies of the content and wall of the FAA were negative. TTE showed good systolic function of both ventricles, normofunctional mitral pros-

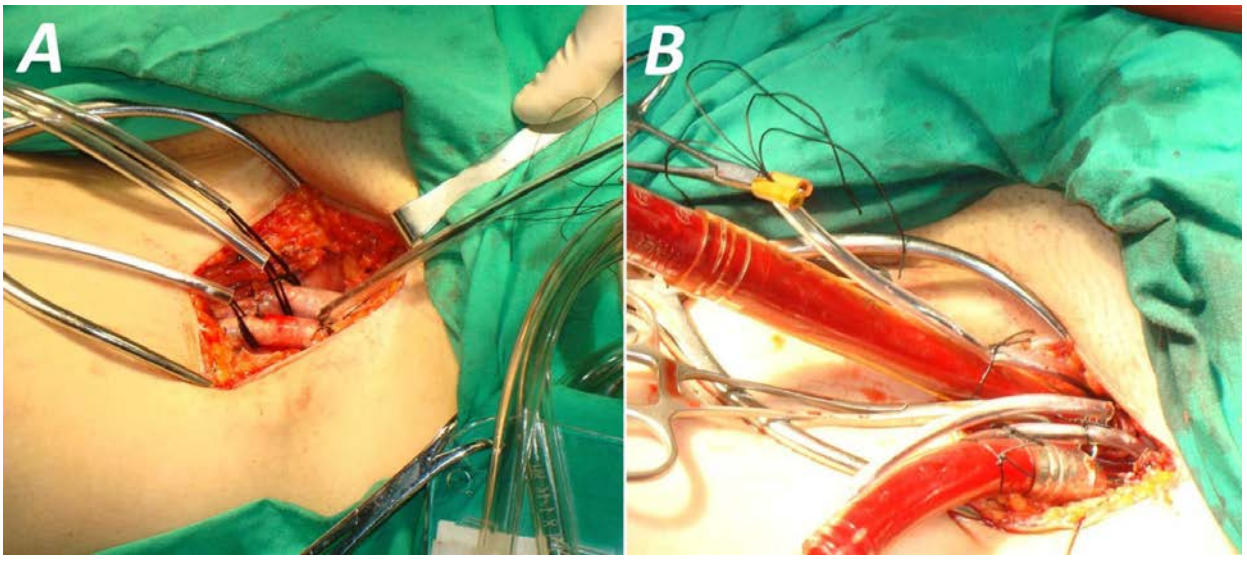

Fig. 2. Peripheral cannulation for cardiopulmonary bypass. **A.** Left femoral artery and vein exposure. **B.** Cannulas placed in both vessels.

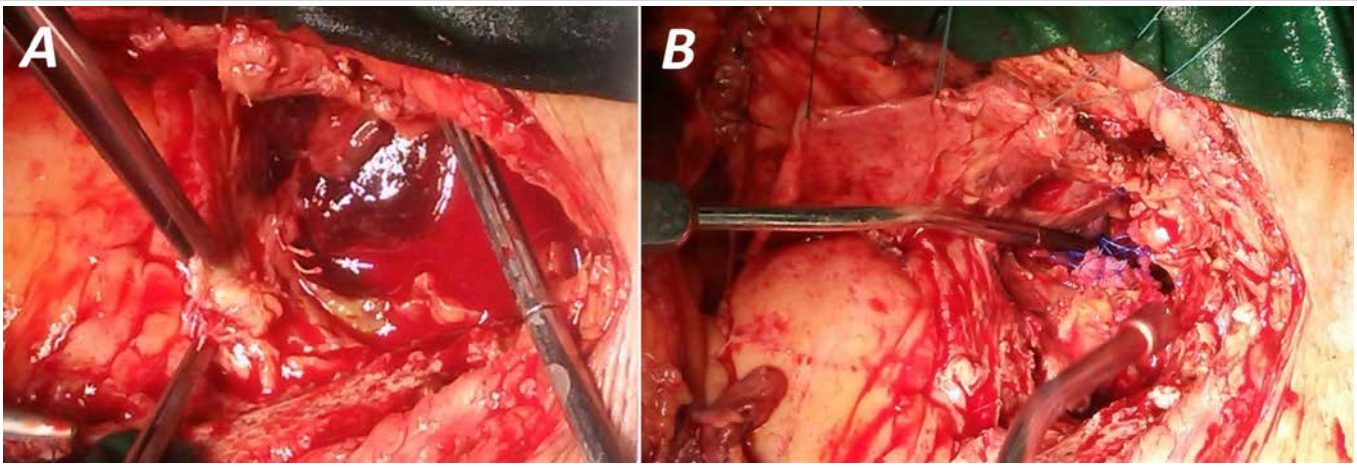

Fig. 3. A. Exposure of the false aneurysm. **B.** False aneurysm repaired with Prolene® 3/0 and Teflon reinforcements.

thesis, with no recurrence of FAA.

COMMENTS

False aneurysms or pseudoaneurysms of the thoracic aorta are an infrequent complication after surgical manipulation of the aforementioned artery, which may appear at the anastomosis or cannulation sites or at the puncture sites for pressure measurements, air purge or injection of cardioplegic solutions. False aneurysms represent a surgical challenge for any work team. Once diagnosed, they are considered surgical emergencies that must be immediately resolved.

Since the late 1990s, researchers have reported acceptable hospital mortality figures, as well as satisfactory mid-term follow-up in patients undergoing surgery due to FAA diagnosed after cardiac surgery[1,2,4-6]. These experiences have made it possible to determine the safest chest reentry strategies[2,7-10].

Its clinical presentation may be acute, threatening the patient's life, as in the case described above; or it may be sub-acute, with non-specific symptoms such as dyspnea, chest pain and fever, among the most mentioned, depending on the series analyzed[4,10,11]; or it may be asymptomatic, diagnosed during follow-up[2,12]. Even insidious forms of presentation accompanied by persistent infection and systemic embolisms have been described[2]. They are diagnosed by imaging procedures[13], where tomography plays a pivotal role for thorough planning of the steps to be followed during surgery by specifying its location, size and relationships with neighboring structures.

Pathologic conditions of the aortic walls, dissections, excessive use of biological glues and infections have been proven to be responsible for the development of FAA after cardiac surgery. Infections are the most feared due to systemic involvement and tissue weakness. Most authors report concomitant infections as the main predisposing factor in a variable number of patients, from 10 to 75%[1,2,4,7,10,14]. The etiological factor is at times difficult to specify, and this great variability is probably conditioned by the different definitions of previous postoperative febrile state, mediastinitis, endocarditis and septic state during reintervention.

The patient presented was in a setting of nosocomial respiratory sepsis after the first surgery that we consider could be expressed in fragility of the tissues and suture lines; although no positive cultures were found later at reintervention. The time lapse between infection and the appearance of FAA is perfectly reasonable. In addition, the rapid growth of the hematoma may have been conditioned by the over-anticoagulation treatment she was receiving (INR figures well above the therapeutic range). Another fact favoring the pathogenic role of infection in the suture line rupture and the appearance of FAA would be the absence of mediastinal widening on X-rays after mitral surgery and normal TTE in follow-up consultations.

False ascending aortic aneurysm may appear secondary to trauma or infection, but prior cardiac surgery is usually the most frequent cause[1]. They have been mostly described after coronary artery bypass graft surgery (in proximal venous graft anas-

tomoses)[15], aortic valve, or surgical procedures related to the ascending aorta such as type A dissections[16]. Initially, aortic cannulation sites to establish CPB were the procedures most associated with the development of these pseudoaneurysms[1], although aortic surgery using prosthetic ducts appears more frequently linked to this complication in the most recent series[2,7]. They have also been described after mitral valve repair[17,18]. Even so, their presentation is usually less than 0.5% of all cases of cardiac surgery[4,7,11,12].

Although uncommon they cause high morbidity and mortality, not only because their rupture could be potentially lethal, but also because of the complex surgical repair interventions required[19]. Its incidence and natural history remain largely unknown. Studies gathering a significant number of patients that could validate the best surgical strategy are scarcely found. There are only isolated case reports[3,20-26] and small heterogeneous series dedicated to describe this unusual complication[1-7,9,10,12,14], where mortality associated with surgical repairs is difficult to determine. The first series described reported a mortality of 20-40%[1,4,9], reaching up to 60% [15]. The most recently published series have managed to reduce it to 6-7%[2,6,7] with the different surgical options implemented.

Conservative treatment does not seem to be the best option in these cases since the small number of patients who have not undergone surgery in some series have very short follow-up periods[5]. Considering its unpredictable course, with fatal rupture, or related sequelae requiring emergency surgery, regardless of the high surgical risk; surgery should be performed as quickly as possible in symptomatic cases. The same behavior should be followed with all diagnosed asymptomatic patients, who must be informed of the implied risks, and with whom the surgical options should be discussed first.

There have been gradual steps in the treatment of this dreaded complication, especially by endovascular approach[27]; but there are certain limitations due to anatomical features and restricted availability in poor countries, being a highly expensive procedure. It is used only for small false aneurysms, with no evidence of sepsis, or signs of compression or ischemia of vital organs. It is especially useful in the elderly or patients at high surgical risk. Literature available reports isolated cases with successful endovascular repair, mainly in the descending aorta[28,29], and some anecdotal in the ascending aorta[30].

Surgery, therefore, continues to play a leading role in most situations we face even in the era of endovascular techniques. Surgical options vary according to the clinical presentation of the FAA, its site and size; and they are often challenging especially in the context of infections, previous cardiac surgery and aortic regurgitation. Most surgical interventions involve the use of CPB and cardioplegic cardiac arrest, or total or partial circulatory arrest, using peripheral sites for arterial and venous cannulation, such as the femoral and subclavian vessels. The experience of each working group and the results of each medical center in using one or another technique are decisive aspects when choosing the surgical option. Femoro-femoral cannulation, with a drop in body temperature to 18°C (64°F), sternal reopening under deep hypothermia and total circulatory arrest is the most commonly used surgical procedure for postoperative false aneurysms[6,14,19].

Two basic aspects are key for successful surgical treatment: preventing surgical accidents resulting from re-entry into the chest, which cause uncontrollable bleeding, and protecting the brain. In considering the first one, we decided to start the CPB with extramediastinal approach before sternal reopening since the FAA was located close to the sternum (less than 2 cm). Most surgical groups agree on this aspect. The question is whether or not to use deep hypothermia[10,14]. This is necessary during circulatory arrest when the location of the FAA prevents aortic clamping and infusion of cardioplegia in the coronary ostiums. Its use implies a series of well-known disadvantages, including imprecise limits for safe-total circulatory arrest-time, and the prolonged CPB time required to cool and then reheat the patient, with its consequent risks of coagulopathies, pulmonary complications and microembolisms. Dodging them, we did not consider lowering the nasopharyngeal temperature before re-entering the mediastinum, since the hole was located in an easily accessible site; and we planned to repair it without cardioplegic arrest with primary sutures; leaving its use reserved in case of complicated false aneurysm approach, or accidental rupture at the point of entrance. Some groups have developed alternatives to reduce long periods of circulatory arrest and deep hypothermia by using a balloon catheter that is inflated through the mouth of the false aneurysm under direct vision, allowing precise placement and avoiding tedious dissections to place the aortic clamp[31]. Axillary artery cannulation combined with femoral cannulation has also been used to insufflate an aortic EndoClamp balloon (remote access perfu-

sion cannula in minimally invasive surgery) and lower body perfusion to prevent hypothermic circulatory arrest[21]. These techniques have critical weaknesses. On the one hand, cannulation of the axillary artery without clamping the origin of the unnamed artery does not reduce the risk of massive bleeding at reopening. On the other hand, the use of an EndoClamp may be dangerous when lacerating or rupturing a fragile aorta, or moving in the arch and occluding any of the supra-aortic vessels.

Bachet *et al*[8] have developed moderate hypothermia with direct cannulation of both carotid arteries approached from the neck (without hindering the surgical field, and easy to clamp) for selective antegrade brain perfusion, and thus treating giant false aneurysms with a safe entry to the mediastinum, which ensures brain protection and avoids the deleterious effects of deep hypothermia. This technique allows dissection of the mediastinum and control of the distal aorta without compromising brain perfusion even in cases of massive bleeding from ruptured FAA, and may influence the quality of complex surgical repair[9]. The evolution of this method, in terms of organ protection, has been published more recently by Martinelli *et al*[32], with the addition of continuous visceral perfusion and ensuring myocardial protection through a catheter in the coronary sinus. A properly inflated balloon catheter is inserted through a femoral artery and, by means of a transesophageal echocardiogram, its position in the descending aorta below the origin of the left subclavian artery is controlled. The other femoral artery and its counterpart vein are cannulated to establish ECC CPB. A percutaneous catheter for retrograde cardioplegia is inserted, under the same echocargiographic control, through the right jugular vein to the coronary sinus to provide the retrograde cardioplegic solution (its position is confirmed with pressure measurements); and both carotid arteries are cannulated to ensure selective cerebral perfusion (as proposed by Bachet *et al*[8]). When body temperature reaches moderate hypothermia (26 to 28 ° C), the ECC CPB is suspended, while selective cerebral perfusion is maintained through both carotid arteries, the cardioplegic solution is administered through the coronary sinus catheter, and at the same time, the balloon is inflated into the descending aorta to initiate selective perfusion of the lower body. This allows for adequate perfusion of the brain, the splenic area and the myocardium; which in turn permits a safely re-entry into the chest, without the use of hypothermic circulatory arrest and without the risk of major bleeding or air embolism, in case of entry into the FAA. Although complex and invasive, this innovative procedure is reserved for high-risk cases and has the disadvantage of not being able to be used in cases of descending aortic disease, especially on dissection.

In patients with complex aortic false aneurysm and aortic valve insufficiency, special attention should be paid to ventricular dilation and subsequent ventricular fibrillation. Transapical venting of the left ventricle through an anterolateral thoracotomy should be used for patients with aortic regurgitation[12].

When addressing the thorax, the most commonly used surgical closure techniques are resection of the FAA and its reconstruction with a pericardium patch or prosthetic material, or simple repairs with direct sutures at the entry site. Patch techniques were widely used since the late 1990s (20th century); however, some groups have dropped them due to recurrence[12]. The replacement of the aortic segment involved in the FAA with a prosthetic duct is the technique of choice for many groups[2,7,9,12] when they face large false aneurysms or graft infections, with or without aortic valve endocarditis.

In the presence of endocarditis, the general principles of resecting and debriding all necrotic tissue are followed, with extensive washing and the use of homografts and biological ducts, associated with prolonged antimicrobial treatment; to avoid recurrences of new FAA. In infections, especially mediastinitis, aggressive treatment should also be prioritized over local or conservative repairs. Coselli *et al*[33] have complemented this strategy with the principle of maintaining circulation "in situ" when facing postoperative infections. For this, they have used additional procedures to cover the aorta, close the dead space and irrigate the mediastinum with antimicrobials. Favorable results have been obtained by mobilizing pericardial fat flaps, muscles and major omentum, and suturing them in the aorta after repair of the FAA. The double-layer autologous patch with fascia lata and saphenous vein has also been used effectively, as an alternative to avoid prosthetic materials in septic false aneurysms[34].

Primary direct closure with simple suture and Teflon supports is another variant of surgical treatment. Considered saving and relatively simple, it may be sufficient in cases where the aortic orifice is small and its location allows it to be used, as happened in this patient in whom the surrounding tissues did not appear septic. Teflon reinforcements

should be anchored into healthy aortic tissue to prevent recurrence. It is necessary to bear in mind that this may be the only appropriate solution in certain patients with high comorbidities, despite the recurrences reported in several publications[4,6,9] that might suggest their abandonment. The goal is to close the neck of the injury, and procedures will be chosen in correspondence with the clinical situation of each particular case.

The interval between the diagnosis of FAA and the initial surgical procedure is variable as evidenced by the various published case series. It may occur within a few days[20], the first 12 months of the postoperative period (where they are quite frequent), after 17 years[9,35], or as far as the third decade after cardiac surgery18. Although infrequent, they should not be ignored. Every surgeon should be aware of the factors invoked in their development and take all the precautionary measures considered necessary to avoid them. They should always be suspected and long-term follow-up with imaging techniques is key at this point. Some authors recommend annual follow-up with contrasted CT scans in patients undergoing aortic surgery, which is more than justified in those with increased risks of developing FAA, with a view to early detection of this complication, which would allow surgical repair at optimal conditions, without deterioration of the patient, and a better prognosis[2,9,12].

CONCLUSIONS

False aneurysm of the ascending aorta is a serious complication of cardiac surgery and is associated with high mortality. Early diagnosis with imaging techniques allows the avoidance of surgical approach in emerging conditions, and improve postoperative outcomes. Sternal reopening is often extremely challenging. Careful preoperative planning is essential for safe re-sternotomy. The combinations of techniques described can help alleviate and minimize blood loss. Proper use of hypothermic circulatory arrest allows dissection of mediastinal structures and aortic control, and methods for preserving cerebral perfusion should be considered. Choosing the peripheral vessel to initiate extracorporeal circulation (femoral, axillary or carotid arteries) depends on the experience of the surgical team and the nature of the particular case. Each case with a diagnosis of false ascending aortic aneurysm should be individualized and treated as such, as the best surgical strategy to avoid perioperative mortality and recurrences is yet to be defined.

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