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Case Series

Surgical management of the complete atrioventricular septal defect: literature review and case report

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ABSTRACT

Complete atrioventricular septal defect is a complex congenital cardiac defect caused by failure of septal separation, frequently associated with Down syndrome. It is considered a significant challenge in terms of diagnostic and therapeutic management. Our work aims to provide an update on the clinical and therapeutic aspects of the complete AVSD managed by our team at University Hospital Ibn Sina of Rabat, highlighting the importance of developing pediatric cardiac surgery in our Moroccan healthcare system. A retrospective descriptive study involved 20 patients who underwent surgery for complete AVSD between January 2020 and December 2023. Medical records were used to collect clinical, echocardiographic, surgical, and follow-up data. The median age at surgery was 7.5 months, and 90% of the patients had Down syndrome. Among the 20 included patients, 19 survived the intervention, yielding a survival rate of 95%. Postoperative complications included transient severe pulmonary hypertension in 4 patients (20%), moderate residual mitral insufficiency in 6 patients (30%), and rare complications such as septic shock and chylothorax. The single-patch repair technique was employed in 95% of cases. Complete CAV requires a multidisciplinary approach. This study highlights the importance of early diagnosis and timely surgical intervention in improving clinical outcomes and reducing morbidity and mortality.

Keywords: Complete atrioventricular septal defect, Congenital heart defect, Down syndrome, Pulmonary hypertension, Left-to-right shunt

INTRODUCTION

The atrioventricular septal defect (AVSD) is a non-cyanogenic congenital heart disease resulting from incomplete development of the separation between the atria and ventricles, accompanied by malformations of the right and left atrioventricular valves.[1]

AVSD accounts for 3% of congenital heart disease.[2] It is usually associated with genetic syndromes, particularly trisomy 21, and represents an important challenge in terms of surgical and medical management. The hallmark anomaly of AVSD is the incomplete development of the atrial septum and associated valves. Several types of AVSD have been described: complete, partial, and intermediate.[3]

In its complete form, the atrioventricular septal defect includes an atrial septal defect of the ostium primum type, a common atrioventricular (AV) valve, and a large ventricular septal defect (VSD) at the ventricular inlet.[4,5] It is often diagnosed early, either before birth by prenatal ultrasound, or during the first months of life due to typical symptoms of left-right shunts such as heart failure, recurrent respiratory infections, and growth retardation.[6]

The management of this malformation relies primarily on surgical correction, which aims to close the septal defects and restore normal valve function. Long-term results vary according to the timing of correction and the presence of complications such as pulmonary hypertension.[7]

CASE SERIES

Patient information

This study includes 20 patients diagnosed with complete AVSD cases operated on by the team of the University Hospital of Rabat between January 2020 and December 2023.

Among the patients, 16 were female (80%) and 4 were male (20%), with a male-to-female sex ratio of 0.25. The mean age of surgical management was 10.75 months, the median was 7.5 months, and the extremes were 4 months and 36 months. Six patients (30%) underwent surgery before the age of 6 months, 8 patients (40%) between 6 and 12 months, 4 (20%) between 12 and 24 months, and 2 (10%) after the age of 24 months (Figure 1).

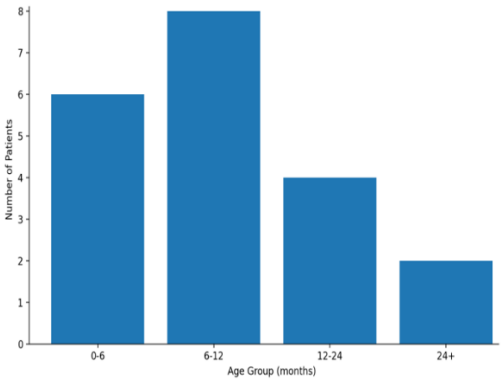

Figure 1: Age distribution at surgery.

Clinical findings

Operated patients had an average weight of 7.21 kg, with a minimum of 4.4 kg and a maximum of 18 kg. The median failure to thrive (weight-for-age) was -3 SD, with a minimum of -4 SD according to growth charts.

All patients presented with preoperative dyspnoea. Recurrent bronchopneumonia was noted in 19 patients (80%). A heart murmur and a B2 pulmonary burst were auscultated in all patients. Clinical signs of trisomy 21 were present in 18 patients (90% of the sample). The diagnosis of trisomy 21 was confirmed by karyotype.

Diagnostic assessment

Chest X-Ray revealed cardiomegaly with vascular overload in all patients (Figure 2). The transthoracic echocardiography showed a common ostium with a mean size of 16±5 mm and a left atrioventricular valve regurgitation: minimal in 8(40%), moderate in 7(35%), and significant in 5(25%). Additional echocardiographic findings included an ostium secondum atrial in 2 patients (10%), a patent ductus arteriosus in 4 patients (20%), left ventricular dilation in 2 patients (10%), and right ventricular dilation in 5 patients (25%) (Table 1).

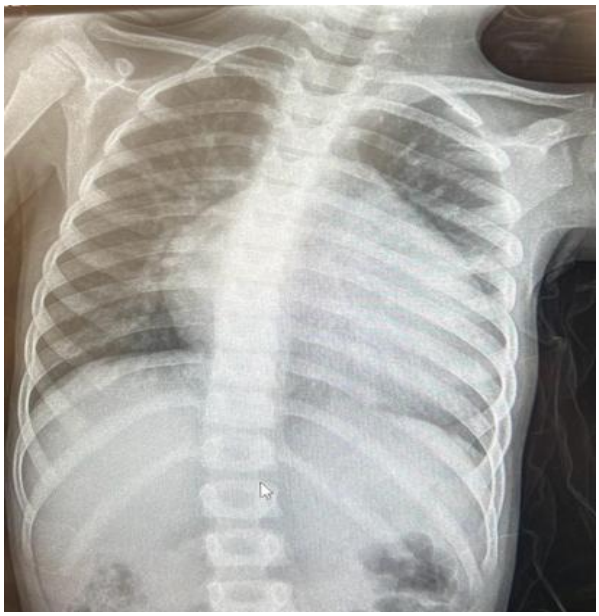

Figure 2: Standard chest X-ray showing cardiomegaly in one of our patients.

Table 1: Preoperative ultrasound data.

Data	N (%)
Commun ostium (mean +/- ET)	16±5 mm
Arterial duct	4 (20%)
Additional CIA	2 (10%)
Left atrioventricular valve regurgitation	
Minimal	8 (40%)
Moderate	7 (35%)
Significant	5 (25%)
Left ventricle	
Dilated	2 (10%)
Non dilated	17 (85%)
Hypoplastic	1 (5%)
Right ventricle	
Dilated	5 (25%)
Non dilated	15 (75%)

Therapeutic intervention

Surgical management primarily involved the single-patch technique, which was utilized in 19 cases (95%), while the Nunn technique was employed in one case (5%). Intraoperative findings revealed the presence of a patent ductus arteriosus in 4 cases (20%), which was surgically ligated. The majority of patients (80%) had type A

AVSD, while 20% had type C AVSD (Figure 3). Cardiopulmonary bypass was required for all patients, with a mean duration of 197 minutes (range: 96–331 minutes). The mean aortic cross-clamp time was 109.8 minutes, with extremes ranging from 11 to 178 minutes (Table 2).

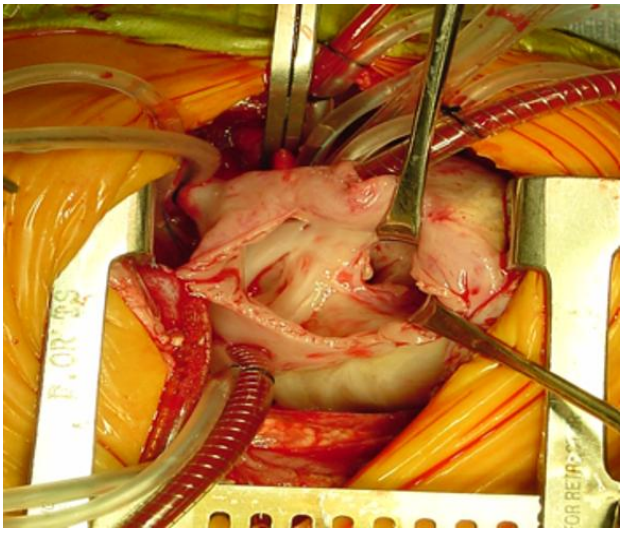

Figure 3: An intraoperative view of a complete atrioventricular septal defect through a right atriotomy.

Table 2: Cardiopulmonary bypass (CPB) data.

Time	Mean (minutes)	Minimum-Maximum (minutes)
Aortic cross-clamp	109,8	11-178
CPB	197	96-331

All patients required invasive mechanical ventilation postoperatively, with a median duration of 3 days. The mean length of stay in the intensive care unit (ICU) was 7±3 days. All patients received adrenaline and milrinone postoperatively, while nine required norepinephrine. To manage pulmonary hypertension, inhaled nitric oxide (NO) was administered to all patients.

Post-operative arrhythmias were observed in 2 patients in our series, presenting with supraventricular tachycardia. None of the patients experienced atrioventricular block. None of the patients required surgical reintervention.

Outcomes

In the short-term postoperative period, 6 patients (30%) developed moderate residual mitral regurgitation, while 4 patients (20%) experienced pulmonary hypertensive crises, which were successfully managed with NO therapy. One patient developed chylothorax, which resolved with appropriate dietary and hygienic measures. Another patient experienced septic shock postoperatively, which was effectively treated with targeted antibiotic therapy.

In terms of medium- and long-term outcomes, 19 patients remained clinically asymptomatic. Echocardiographic follow-up revealed no residual shunt in 14 patients, while 7 patients had minimal shunting. One patient developed left heart chamber dilation. Left atrioventricular valve regurgitation was observed in 15 patients (75%) at a mild grade and in 5 patients (25%) at a moderate grade. Unfortunately, one patient succumbed to complications due to an unbalanced AVSD associated with left ventricular hypoplasia (Table 3).

Table 3: Postoperative echocardiography data.

Data	N (%)
Residual shunt	
Absent	14 (70)
Minimal	7 (30)
Dilated left heart chambers	1 (5)
Left atrioventricular valve regurgitation	
Mild	15 (75)
Moderate	5 (25)

DISCUSSION

Complete atrioventricular septal defect is a complex and life-threatening congenital heart defect, associated with multiple comorbidities, notably a high prevalence of Down syndrome.[8] This condition presents a real challenge for cardiac surgery and pediatric cardiology teams worldwide: newborns with Down syndrome are at an increased risk of developing neonatal pulmonary hypertension. This is likely due to structural vascular abnormalities, impaired nitric oxide production, an excess of anti-angiogenic factors, and pulmonary hypoplasia.[9] In our series, data reveal a higher proportion of Down syndrome compared to those reported by the literature: 18 out of 20 cases.[10,11]

The age at surgery plays a critical role in postoperative outcomes, affecting both mortality and the risk of complications. Crawford reported an average surgical age of 5 months in a case series, with extremes of 1.3 to 15.1 months and an average weight of 4.8 kg (extremes of 2.1 to 7.3 kg).[12] Soliman et al noted that the age at surgery ranged from 4 to 47 months, with a median of 8 months, suggesting that surgery can be safely performed slightly later in some cases.[13] In contrast, St. Louis et al observed a median surgical age of 4.6 months and a median weight of 5.0 ± 1 kg.[14] However, infants operated on before 2.5 months had less favorable outcomes, with increased in-hospital mortality (9.5%), a higher incidence of major complications, and prolonged postoperative stays. Although early intervention is generally recommended to prevent complications such as heart failure and pulmonary hypertension, St. Louis advises performing AVSD repair between 3 and 6 months of age when

feasible.[14] This timing balances the need to prevent irreversible pulmonary vascular changes by minimizing the risks associated with surgery in very young infants. This strategy aims to optimize outcomes by reducing mortality and complication rates while preventing the progression of pulmonary vascular disease.

In our case series, the average age for surgical management was 10.75 months, with a median of 7.5 months and extremes ranging from 4 to 36 months.

This surgical age is justified because pulmonary arterial hypertension caused by left-to-right shunts in children with Down syndrome is likely to become irreversible earlier than in non-syndromic children, as early as six months of age. There is a clear benefit, as shown, to setting the age for complete AVSD surgery at six months rather than one year.[15]

Early surgery impacts not only pulmonary circulation and complications related to pulmonary hypertension but also the competence of the left atrioventricular valve. Volume overload and valvular regurgitation contribute to progressive annular dilation, worsening leaflet coaptation. This process is accompanied by leaflet dysplasia in the apposition area, further affecting coaptation mechanisms. This "regurgitation begets regurgitation" cycle is interrupted early with timely intervention.

A counterargument to early repair is the risk of suture dehiscence in the apposition zone in patients weighing less than 5 kg at the time of surgery, but this is debatable.[16]

In our case series, six patients (30% of the sample) underwent surgery before six months of age, eight (40%) between six and twelve months, four (20%) between twelve and twenty-four months, and two (10%) at an age exceeding twenty-four months.

Delays in management can often be attributed to late diagnosis, highlighting the importance of training autonomous specialists in this field and integrating congenital heart disease awareness into basic medical education. Financial constraints and delays in obtaining financial support also contribute to these delays.

There is a lack of bibliographic data and guidelines regarding the management and operability of children presenting with advanced stages of congenital heart disease.[17] The majority of these patients come from countries or regions where despite various strategies, surgical interventions cannot always be performed within recommended timelines.[18]

A "late form" of complete AVSD is defined when the patient's age exceeds six months. Several factors contribute to late presentation: delayed diagnosis, lack of resources and infrastructure, insufficient awareness, and inadequate medical advice.

The management of these patients is complex, particularly due to the assessment of operability, which remains difficult in the absence of consensus on echographic variables and those measured by cardiac catheterization. The latter is necessary in the absence of signs of pulmonary overload, bidirectional shunting, the presence of post-tricuspid communication, or low systemic oxygen saturation values. A pulmonary resistance value of 4 Wood units/m², as well as an RV:SV ratio below 0.3, have been proposed as thresholds for considering surgery. Delayed surgery is characterized by a post-operative period marked by a state of severe low cardiac output, resulting from dysfunction of the systemic or biventricular ventricle. This situation requires specific management and a highly specialized approach. Surgical considerations also vary in these patients and may include: delayed sternotomy closure due to the often-present high pulmonary hypertension, the early implantation of a peritoneal dialysis catheter due to the risk of low cardiac output, and the maintenance of a patent foramen ovale for decompression.[17]

In our case series, the management of AVSDs operated on at an advanced age is based on a strategy combining surgical interventions and medical treatments, following international recommendations, while relying on local experience due to the lack of specific bibliographic data on this subject. Keeping the chest open is generally not required, but a patent foramen ovale is often preserved in patients at risk for pulmonary hypertension.

Medically, management includes normalizing calcium levels, administering physiological doses of hydrocortisone, and treating hypothyroidism, which is commonly observed in patients with Down syndrome. It also addresses other comorbidities associated with this condition, including ENT, respiratory, and digestive issues.

Reducing metabolic needs relies on effective analgesia, appropriate sedation, and muscle relaxation through curarization, accompanied by optimized mechanical ventilation and strict temperature control. Managing pulmonary hypertension crises primarily focuses on eliminating triggering factors such as pain, agitation, hypercapnia, and acidosis. Milrinone, nitric oxide, and sildenafil are key elements in this management.[19,20]

Recurrent bronchopulmonary disease has been observed in all the patients in our series. Respiratory infections are one of the main causes of morbidity and mortality in children under 5 years old, with respiratory syncytial virus being the most frequently implicated agent.[21] Increased pulmonary vascular resistance, commonly found in congenital heart diseases, is one of the factors exacerbating the progression of these infections.[22] This may be explained by airway compression due to

pulmonary artery dilation, cardiac chamber dilation, and pulmonary hyperflow, which leads to an increased risk of pulmonary overload and reduced alveolar reserve. These respiratory complications result in pulmonary sequelae that complicate post-operative respiratory management, with a prolonged need for invasive ventilation and the necessity of non-invasive ventilation, thus extending the duration of hospitalization in the intensive care unit.

The need for reoperations after the initial AVSD repair is a reality for several patients, often due to recurring issues such as severe left atrioventricular (AV) valve regurgitation and other structural complications. According to Crawford, about 10.2% of patients required reoperation due to severe mitral regurgitation.[12] In Daene's study, 4.5% of patients required reoperation within 30 days of the initial repair, largely due to complications like left AV valve regurgitation and subaortic stenosis.[23] In Soliman's study, reinterventions were necessary in 6.2% of cases, often due to valve regurgitation or insufficient correction of a residual shunt.[13]

In our series, the reoperation rate is zero. These optimistic figures are likely due to the limited follow-up, which averages just over two years.

The postoperative length of stay after complete AVSD repair is influenced by several clinical factors, particularly the patient’s weight and age at the time of surgery. In St Louis' study, the median postoperative stay was 8 days.[14] However, low-birth-weight infants and those operated on at a very young age had prolonged stays due to increased complications and age-related fragility.

In our case, the average length of stay is 7 days. These data emphasize the importance of considering age and weight in planning postoperative care, particularly for younger infants and low-birth-weight patients, who may require more intensive monitoring to ensure optimal recovery.

In Crawford's study, the in-hospital mortality rate after surgical AVSD repair was 3%, but it varied according to the patient's age and weight.[12] Mortality reached 15.2% in patients weighing less than 3.5 kg and 9.5% in those operated on before 2.5 months of age. Atz et al report similar results, with a hospital mortality rate of 2.5% and a tendency for increased risk in surgeries before 2.5 months (12% mortality at six months compared to 2% after this age).[24] These data highlight that low weight and early age are major risk factors, associated with increased fragility and surgical complexity.

In our series, one death was registered (5%), due to an anatomical subtype discovered during the intervention: unbalanced complete atrioventricular septal defect with left ventricular hypoplasia.

Concerning the surgical technique, both double-patch and single-patch approaches are commonly used. The choice depends on the anatomy and complexity of the case: Soliman favors the double-patch technique for its anatomical precision and stability, while Crawford highlights the growing popularity of the single-patch technique, associated with zero mortality rates in his recent series.[13,12] Malik et al confirm that the single-patch technique, which is simpler and quicker, is suitable for less complex cases, while the double-patch is preferred for more complex repairs.[25]

Finally, our case series favors the single-patch technique, used in the majority of patients to minimize residual shunts and atrioventricular blocks.

The average aortic cross-clamping time is 109.8 minutes, which is higher compared to the various series described: 76 minutes in Fong's series and 104 minutes in Prifti's series.[26,27]

Postoperative complications following AVSD repair vary in severity and frequency, but they are influenced by several factors, including age, weight, and pre-existing comorbidities.

In Crawford's study, the incidence of complications such as complete heart block was relatively low, with only 3.4% of patients.[12] However, mitral valve regurgitation remains a recurrent problem, with about 7 to 8% of patients needing re-intervention. Daene's study highlights other complications, such as pleural effusion, respiratory infections, and residual mitral insufficiency.[23] Arrhythmias were observed in 6.7% of cases, and 8.2% of patients developed heart failure during follow-up, indicating potentially serious sequelae even after successful repair. St Louis' analysis reports major complications in 9.8% of cases, including renal failure, neurological deficits, and arrhythmias requiring a permanent pacemaker (2.7% of patients).[14] Finally, according to Malik, certain preoperative factors such as left AV valve regurgitation and significant pulmonary hypertension are associated with poorer outcomes.[25] Myocardial protection during surgery and careful management of postoperative pulmonary hypertensive crises are therefore essential to improve long-term survival chances and reduce the need for reinterventions.

In our series, our results align with the literature, with minimal medium-term postoperative complications. Postoperative follow-up was marked by the onset of severe pulmonary arterial hypertension requiring sedation, assisted ventilation, and nitric oxide (NO) administration in 4 patients. In all cases, this pulmonary hypertension was reversible; the early age of surgery allowed for rapid relief of the pulmonary arterioles from the high pressures caused by congenital heart disease, leading to a reduction in vascular resistance. This is also confirmed by other studies. Additionally, 6 patients, had moderate (grade II) residual mitral insufficiency, while 2

patients suffered from supraventricular tachycardia. One case of chylothorax and one case of septic shock were also observed.

Infectious complications are an integral part of surgery and increase the difficulty of management due to the rising cost of care, prolonged hospitalization, and mortality.[28] Surgical site infections (SSIs) are among the most common nosocomial infections, with an estimated incidence ranging from 2% to 11% for all surgical procedures.[29] In cardiac surgery, surgical site infection is estimated at 3.6%, according to A.F. Tejerina, depending on risk factors, compliance with preventive measures, and the rigor of monitoring.[30] The skin of the thoracic region, due to its abundance of sebaceous glands, provides a favorable environment for the growth of lipophilic microorganisms such as Propionibacterium spp. and Malassezia spp.[31] Moreover, Staphylococcus aureus is now recognized as the predominant pathogen in surgical site infections (SSIs), as emphasized by P. Zarb.[32]

The prevention of SSIs relies on targeted actions implemented at each stage of care: preoperative, intraoperative, and postoperative.[32] In our team, particular attention is given to the implementation of these measures, which include the assessment of nutritional status and adjustment of management in cases of malnutrition, which is common in patients with AVSD, particularly in late forms. Other key measures include the administration of broad-spectrum antibiotic prophylaxis and rigorous hand hygiene using antiseptic solutions based on chlorhexidine.

Congenital heart diseases promote the development of infective endocarditis due to various factors, including the shear forces exerted on the endothelium, the presence of intracardiac devices (such as valves, patches, or prosthetic conduits), implanted electronic devices, and certain medical procedures.[33] Published studies show that endothelialization of prosthetic devices implanted during cardiac surgery is generally completed within six months after the operation. During this period, microorganisms from medical procedures or daily activities may adhere more readily to the repaired sites. Furthermore, it is plausible that bacteremias, directly caused by the surgical procedure, are involved in the development of infective endocarditis.[34]

Limitations

Being hospital-based, it introduces a selection bias, excluding children with sub-clinical symptoms or with no access to information. Although the study results are interesting, the small sample size restricts the external validity of the conclusions. The retrospective nature of the study was limited by missing data. Long-term follow-up of the data from our series will require in-depth analysis in the coming years when clinical follow-up becomes more substantial.

CONCLUSION

Complete atrioventricular canal is a rare and complex congenital heart disease that holds significant clinical importance, especially in the context of genetic syndromes such as trisomy 21. Early diagnosis, facilitated by echocardiography is crucial to reduce morbidity. Management requires a multidisciplinary approach, with surgery being the cornerstone of treatment. Our study describes the clinical and therapeutic profile of patients followed by our team, highlighting a strong association with trisomy 21 and advanced age at the time of surgery which complicates care due to associated conditions, such as repeated respiratory infections and malnutrition. Long-term follow-up is essential to prevent complications such as heart failure and pulmonary hypertension. Advancements in surgery and pathophysiological understanding will further improve prognosis. Despite these challenges, the surgical outcomes of our study are comparable to international standards, and provide valuable insights into the challenges of managing pediatric heart disease within the national healthcare system.

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