International Journal of Medical Science and Clinical Research Studies

ISSN(print): 2767-8326, ISSN(online): 2767-8342

Volume 04 Issue 11 November 2024

Page No: 2173- 2179

DOI: https://doi.org/10.47191/ijmscrs/v4-i11-36, Impact Factor: 7.949

Comprehensive Insights into Cor Triatriatum: An Overview of Pathophysiology, Clinical Presentation, Diagnostic Strategies, and Management Options

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ABSTRACT

Cor triatriatum is a rare congenital cardiac malformation characterized by the presence of a fibromuscular membrane that divides one of the atria into two distinct chambers, impeding normal blood flow. It is typically classified into Cor triatriatum sinister, affecting the left atrium, and Cor triatriatum dexter, involving the right atrium. The condition arises from aberrant embryologic development, with a spectrum of clinical manifestations ranging from asymptomatic cases to severe heart failure or cardiogenic shock in neonates and infants. Advances in diagnostic imaging, particularly echocardiography and cardiac magnetic resonance imaging (MRI), have improved our ability to identify and characterize this anomaly. Surgical correction remains the definitive treatment, though minimally invasive techniques are emerging as viable options for selected patients. This article aims to provide a detailed review of the etiology, anatomical variations, pathophysiological impact, diagnostic approach, and both traditional and novel management strategies for Cor triatriatum. Additionally, the article discusses prognostic factors and the role of long-term follow-up in optimizing patient outcomes.

KEYWORDS: Cor triatriatum, congenital heart disease, cardiac embryology, atrial septation, echocardiography, cardiac MRI, surgical correction, heart failure, pediatric cardiology

ARTICLE DETAILS

Published On:
26 November 2024

Available on:
https://ijmscr.org/

INTRODUCTION

Cor triatriatum represents a congenital anomaly of the heart in which a membrane divides either the left or right atrium into two distinct chambers, significantly disrupting hemodynamic flow and creating a clinical picture that can vary dramatically depending on the degree of obstruction and patient age. The malformation was first described in the 19th century, and despite advances in our understanding, it remains a challenging diagnosis due to its rarity and variable presentation.1,2

Embryologically, Cor triatriatum sinister arises from incomplete incorporation of the common pulmonary vein into the left atrium, leading to a partition that separates the pulmonary venous return from the mitral valve. Cor triatriatum dexter, far less common, results from abnormal persistence of the right sinus venosus valve. These developmental anomalies culminate in pathophysiological consequences that can manifest as pulmonary venous hypertension, atrial arrhythmias, or heart failure, particularly when associated with additional cardiac malformations, such as atrial septal defects.1.2

The clinical course of Cor triatriatum is highly variable. While some patients remain asymptomatic for years, others, especially infants, may present with severe respiratory distress and failure to thrive. In adults, symptoms often mimic those of mitral stenosis, including dyspnea, orthopnea, and reduced exercise tolerance. Accurate and timely diagnosis is paramount, with echocardiography serving as the primary

tool, though cardiac MRI and computed tomography (CT) may be necessary for detailed anatomical assessment.2,3

Management strategies are tailored to symptom severity and anatomical findings. Surgical resection of the membrane, historically the gold standard, has a high success rate, while minimally invasive approaches, such as percutaneous interventions, are gaining traction for select cases. Long-term outcomes generally depend on the promptness of diagnosis and treatment, with early intervention being crucial in preventing irreversible complications. This review seeks to elucidate the complexities of Cor triatriatum, emphasizing a comprehensive, multidisciplinary approach to care.2,3

EPIDEMIOLOGY

Cor triatriatum is a rare congenital cardiac anomaly with a reported prevalence of approximately 0.1% to 0.4% among congenital heart defects, which themselves occur in roughly 8 per 1,000 live births. This malformation is more commonly observed in the left atrium, termed Cor triatriatum sinister, with Cor triatriatum dexter being significantly less frequent. The condition does not exhibit a clear sex predilection, although some studies have suggested a slight male predominance. The incidence of Cor triatriatum varies globally, but large-scale epidemiological data are limited due to its rarity and the tendency for cases to be identified primarily at tertiary care centers with specialized pediatric and cardiology services.2,3

Most cases of Cor triatriatum are detected during infancy or childhood, particularly when the membranous division causes severe hemodynamic compromise. However, a substantial number of individuals with less obstructive forms may remain undiagnosed until adulthood, often presenting with symptoms that mimic mitral valve pathology or are found incidentally during imaging for unrelated conditions. Advances in diagnostic imaging modalities, especially echocardiography, have enhanced the detection rates of Cor triatriatum, even in asymptomatic or mildly symptomatic individuals.2,3

Congenital heart defects are commonly associated with Cor triatriatum. Among these, atrial septal defects (ASDs) are frequently reported and are present in approximately 25% to 80% of cases. Other congenital cardiac anomalies that may coexist include persistent left superior vena cava, anomalous pulmonary venous connections, and complex syndromic presentations involving both cardiac and extracardiac abnormalities. These associations complicate the hemodynamic impact and clinical management of Cor triatriatum and may influence the timing and approach to intervention.3,4

The survival and clinical course of patients with Cor triatriatum are significantly influenced by the degree of obstruction and the presence of associated anomalies. Historically, the condition was often fatal in early life due to severe congestive heart failure or pulmonary venous hypertension. However, with advancements in surgical techniques and neonatal intensive care, survival rates have improved dramatically. Despite this, long-term epidemiological studies on the outcomes and quality of life in patients with corrected Cor triatriatum are limited, and there remains a need for comprehensive registries to better understand the natural history and prognostic factors associated with this anomaly.4,5

Furthermore, the advent of fetal echocardiography has made prenatal diagnosis increasingly feasible, allowing for early detection and perinatal planning, although the impact of such early identification on overall prevalence and outcomes remains to be fully elucidated. Population-based studies are needed to gain a clearer understanding of the true prevalence and demographic variations of Cor triatriatum, as well as to investigate potential genetic and environmental risk factors that may contribute to its development.4,5

CLINICAL MANIFESTATIONS

The clinical presentation of Cor triatriatum is highly variable and depends on the degree of obstruction caused by the fibromuscular membrane dividing the atrium, the age at presentation, and the presence of associated cardiac anomalies. The symptomatology arises from impaired blood flow, leading to elevated pulmonary venous pressures, pulmonary congestion, and, in severe cases, heart failure.4,5

Neonatal and Pediatric Presentation:

In neonates and infants, Cor triatriatum sinister can present with profound symptoms, particularly when the membrane severely restricts pulmonary venous outflow. Infants may exhibit signs of congestive heart failure, including tachypnea, dyspnea, poor feeding, failure to thrive, and significant respiratory distress. Severe cases may result in cyanosis and life-threatening pulmonary edema, especially when associated with additional cardiac defects that exacerbate the hemodynamic burden. Pulmonary hypertension is a frequent consequence of significant obstruction, further complicating the clinical picture.4,5

Recurrent respiratory infections are common among pediatric patients, often prompting an initial evaluation that leads to the diagnosis. Some children may also display exercise intolerance or fatigue due to reduced cardiac output. In cases where the membranous division causes only partial obstruction, symptoms may be more insidious, delaying diagnosis until later in childhood or adolescence.6

Adult Presentation:

In adults, Cor triatriatum is often diagnosed incidentally or during an evaluation for unexplained dyspnea or symptoms mimicking mitral valve stenosis. These patients may experience progressive exertional dyspnea, orthopnea, paroxysmal nocturnal dyspnea, palpitations, and chest discomfort. Symptoms result from increased left atrial pressure and pulmonary venous hypertension. Adults with

undiagnosed or minimally obstructive Cor triatriatum can present later in life with atrial arrhythmias, most commonly atrial fibrillation or atrial flutter, secondary to chronic atrial dilation and pressure overload. Hemoptysis and thromboembolic events may occur in patients with long-standing pulmonary venous hypertension and atrial dilation.6

Signs and Examination Findings:

Physical examination findings are non-specific and vary with the severity of the obstruction. In severe cases, a precordial bulge, palpable heave, or a loud P2 component of the second heart sound may be appreciated, indicative of pulmonary hypertension. Auscultatory findings may include a diastolic rumble at the apex, resembling that of mitral stenosis, or signs of right heart failure, such as jugular venous distention, hepatomegaly, and peripheral edema, in cases of severe pulmonary venous hypertension and right-sided heart strain.6 Mild cases may have no discernible physical findings, while severe obstruction can lead to a clinical picture of cardiogenic shock or cyanosis in neonates. The presence of associated congenital defects, such as atrial septal defects (ASDs), can further complicate the clinical presentation. For example, a large ASD may serve as a pressure-relief mechanism, altering the expected hemodynamic and clinical signs.7

Complications and Associated Conditions:

Cor triatriatum is frequently associated with other structural heart abnormalities, such as ASDs, patent foramen ovale, persistent left superior vena cava, and anomalous pulmonary venous return. These associations can mask or accentuate symptoms and necessitate a comprehensive echocardiographic evaluation to delineate the complete anatomic picture. Complications of Cor triatriatum include pulmonary venous hypertension, atrial arrhythmias, systemic thromboembolism, and progressive right ventricular failure. The chronic elevation in pulmonary pressures may lead to irreversible pulmonary vascular remodeling and right heart failure if left untreated.7

Differences Between Cor Triatriatum Sinister and Dexter:

While Cor triatriatum sinister, involving the left atrium, is far more common and presents with symptoms related to pulmonary venous obstruction, Cor triatriatum dexter, involving the right atrium, leads to right-sided heart failure manifestations. Cor triatriatum dexter often presents with signs of systemic venous congestion, including hepatomegaly, ascites, and lower extremity edema, particularly when severe.7

In summary, the clinical manifestations of Cor triatriatum encompass a wide spectrum, ranging from asymptomatic cases discovered incidentally to life-threatening presentations in infants. The hemodynamic impact of the atrial membrane, the extent of associated cardiac anomalies, and the progression of secondary pulmonary hypertension collectively determine the clinical course. Early recognition and timely intervention are critical in preventing irreversible sequelae and optimizing long-term outcomes.7

DIAGNOSTIC METHODS

The diagnosis of Cor triatriatum requires a comprehensive assessment involving a combination of clinical evaluation, advanced imaging modalities, and, in some cases, cardiac catheterization to fully delineate the anatomical and hemodynamic complexities associated with this congenital anomaly. Early and accurate diagnosis is essential for optimizing management and preventing complications, particularly in cases where the obstructive membrane significantly impacts cardiac function and pulmonary hemodynamics.8

Clinical Evaluation and Initial Assessment:

The initial suspicion of Cor triatriatum is often raised based on clinical history and physical examination findings suggestive of elevated pulmonary venous pressure or heart failure. In infants and young children, symptoms such as respiratory distress, poor feeding, and failure to thrive may prompt further investigation, while adults presenting with dyspnea, fatigue, or signs mimicking mitral stenosis require detailed evaluation. Auscultatory findings, including a diastolic murmur at the apex, may guide clinicians to suspect a structural cardiac abnormality.8

Echocardiography:

Transthoracic echocardiography (TTE) serves as the cornerstone of diagnostic evaluation for Cor triatriatum and is typically the first-line imaging modality. TTE provides essential information regarding the presence, location, and characteristics of the atrial membrane dividing the left or right atrium. Two-dimensional (2D) imaging allows visualization of the fibromuscular partition, and Doppler echocardiography can assess the hemodynamic impact of the obstruction, including gradients across the membrane and evidence of elevated pulmonary pressures.8

For detailed anatomical assessment, transesophageal echocardiography (TEE) may be required, especially in cases where the transthoracic windows are suboptimal or when a more precise delineation of the atrial anatomy is necessary. TEE offers superior resolution of the interatrial septum, the membranous division, and associated structures, making it invaluable for preoperative planning. Color Doppler imaging can further elucidate abnormal flow patterns, quantify the degree of obstruction, and evaluate for associated intracardiac defects, such as atrial septal defects (ASDs) or anomalous pulmonary venous connections.8

Cardiac Magnetic Resonance Imaging (MRI):

Cardiac MRI is an advanced imaging modality that provides comprehensive three-dimensional visualization of cardiac structures, offering unparalleled detail of the atrial anatomy and membranous partition. MRI is particularly useful for assessing the relationship between the obstructive membrane

and the pulmonary veins, as well as for evaluating extracardiac structures. Additionally, cardiac MRI can quantify chamber volumes, assess myocardial function, and provide accurate measurements of flow gradients across the membrane. This modality is often employed when echocardiographic findings are inconclusive or when a more detailed anatomical and functional assessment is required for surgical planning.8

Computed Tomography (CT) Angiography:

CT angiography is another valuable tool for visualizing the intricate anatomy of Cor triatriatum, especially in cases where there is suspicion of complex or anomalous pulmonary venous return. High-resolution, contrast-enhanced CT can delineate the fibromuscular membrane and its impact on the atrium and pulmonary veins. It also provides critical information on associated vascular and extracardiac abnormalities. CT imaging is particularly beneficial for patients who cannot undergo MRI or for those requiring detailed anatomical mapping prior to surgical or interventional procedures.8

Cardiac Catheterization:

While non-invasive imaging modalities are generally sufficient for diagnosis, cardiac catheterization may be necessary in select cases to provide hemodynamic data and assess the severity of pulmonary hypertension. Cardiac catheterization can measure pressure gradients across the atrial membrane and evaluate pulmonary artery pressures, which are crucial for determining the hemodynamic significance of the obstruction. Additionally, angiography performed during catheterization can offer further insight into the anatomy of the pulmonary veins and the extent of obstruction. This invasive approach is typically reserved for cases in which non-invasive imaging does not provide definitive information or when interventional planning is required.8

Electrocardiography (ECG) and Chest Radiography:

Electrocardiography may reveal non-specific findings, such as left atrial enlargement, right ventricular hypertrophy, or atrial arrhythmias, depending on the severity of the obstruction and the degree of pulmonary hypertension. Although not diagnostic, ECG findings can support the clinical suspicion of a structural cardiac anomaly. Chest radiography may show signs of pulmonary congestion, cardiomegaly, or prominent pulmonary arteries indicative of pulmonary venous hypertension. These findings are often non-specific but can be suggestive of underlying cardiac disease, prompting further evaluation with advanced imaging techniques.9

Differential Diagnosis:

It is essential to distinguish Cor triatriatum from other conditions that present with similar clinical and hemodynamic features, such as mitral stenosis, supramitral ring, and pulmonary vein stenosis. Echocardiographic and advanced imaging modalities are critical for differentiating these conditions and confirming the diagnosis of Cor triatriatum. In complex cases, a multidisciplinary approach involving pediatric cardiologists, radiologists, and cardiothoracic surgeons is often required to ensure accurate diagnosis and optimal management.9

In summary, the diagnosis of Cor triatriatum relies on a meticulous approach utilizing a combination of clinical assessment and advanced imaging techniques. Echocardiography remains the mainstay of diagnosis, while cardiac MRI and CT provide detailed anatomical information. Cardiac catheterization serves a complementary role in cases requiring precise hemodynamic evaluation. Accurate and early diagnosis is pivotal in guiding appropriate therapeutic interventions and improving patient outcomes.9

CURRENT TREATMENT OPTIONS

The management of Cor triatriatum is highly individualized and depends on the severity of the hemodynamic obstruction, the presence of symptoms, and any associated congenital heart defects. Treatment strategies have evolved significantly, with the goal of restoring normal blood flow, alleviating pulmonary venous hypertension, and preventing long-term complications. Management typically involves a combination of surgical and, in select cases, catheter-based interventions.9

Surgical Treatment:

Surgical correction remains the gold standard for the management of symptomatic Cor triatriatum or in cases where the membranous partition significantly impedes blood flow. The primary objective of surgery is to excise or fenestrate the fibromuscular membrane dividing the atrium, thereby restoring unobstructed blood flow from the pulmonary veins into the left atrium or from the systemic veins into the right atrium, depending on whether Cor triatriatum sinister or dexter is present.9

The surgical approach is generally performed via a median sternotomy with cardiopulmonary bypass. After careful inspection of the atrial anatomy, complete resection of the membrane is undertaken. In cases where the membrane is closely adherent to surrounding structures or if there is a risk of damaging vital cardiac components, fenestration or partial excision may be performed to alleviate the obstruction. Special attention is paid to preserving the integrity of the atrioventricular valves and ensuring that all pulmonary veins have an unobstructed pathway into the atrium. The surgical outcomes are typically excellent, with low morbidity and mortality rates, especially when performed in specialized centers by experienced pediatric or congenital heart surgeons.10

For patients with associated cardiac anomalies, such as atrial septal defects (ASDs), the surgical procedure may include simultaneous repair of these defects. In cases where there are

complex congenital anomalies or if additional surgical interventions are needed, a comprehensive surgical plan is developed to address all anatomical abnormalities in a single operative session. Surgical repair in the neonatal or early infancy period is crucial for patients presenting with severe symptoms or heart failure, as delayed intervention can lead to irreversible pulmonary vascular changes and long-term complications.11

Minimally Invasive and Catheter-Based Interventions:

Although surgical resection remains the primary treatment modality, advances in interventional cardiology have paved the way for minimally invasive approaches in selected cases. Catheter-based interventions, such as balloon dilation or transcatheter membrane fenestration, are being explored as alternative options, particularly for patients who are not ideal surgical candidates due to comorbidities or high surgical risk.11

Balloon dilation can be used to create a controlled rupture or fenestration of the membrane, thereby reducing the pressure gradient and improving hemodynamics. However, this approach is generally reserved for patients with milder forms of obstruction or as a palliative measure in critically ill neonates who require immediate relief from severe obstruction but are not yet suitable for definitive surgical repair. The long-term efficacy of these catheter-based interventions remains a topic of ongoing research, and outcomes are variable depending on the anatomical characteristics of the membrane and the presence of other cardiac anomalies.12

Medical Management:

While medical therapy is not curative for Cor triatriatum, it plays a supportive role in managing symptoms and stabilizing patients before definitive surgical or interventional treatment. Diuretics, such as furosemide, are commonly used to manage pulmonary congestion and heart failure symptoms by reducing preload and alleviating pulmonary venous hypertension. In patients with significant atrial arrhythmias, antiarrhythmic medications may be employed to control heart rate and rhythm. Beta-blockers or calcium channel blockers can be used to manage tachyarrhythmias, although their use is guided by the clinical presentation and hemodynamic status of the patient.12

Pulmonary vasodilators, such as sildenafil or bosentan, may be considered in patients with severe pulmonary hypertension secondary to prolonged obstruction. These agents help to lower pulmonary artery pressures and improve cardiac output. However, their use should be carefully monitored, and they are typically considered as an adjunct to surgical intervention rather than as a standalone therapy. For neonates and infants presenting with acute heart failure, inotropic support and mechanical ventilation may be required as temporizing measures while planning for surgical correction.12

Postoperative Care and Long-Term Follow-Up:

Postoperative management focuses on monitoring for complications such as residual obstruction, atrial arrhythmias, or pulmonary hypertensive crises. Echocardiographic assessment is routinely performed to ensure that the surgical repair has successfully alleviated the obstruction and to evaluate for any residual or recurrent membrane formation. Long-term follow-up is essential, particularly in patients who required early intervention or had significant pulmonary hypertension prior to surgery. Surveillance echocardiography and periodic assessment of exercise tolerance are recommended to monitor cardiac function and pulmonary pressures.13

Patients who undergo successful surgical correction generally have an excellent prognosis, with significant improvement in symptoms and quality of life. However, there is a risk of developing atrial arrhythmias later in life, particularly in those who presented with significant atrial dilation or fibrosis. Lifelong cardiology follow-up is advised to monitor for late complications and to ensure optimal management of any residual or evolving cardiac issues.14

Emerging Therapies and Future Directions:

Ongoing research is focused on improving the outcomes of patients with Cor triatriatum through the development of novel surgical techniques, better risk stratification tools, and advancements in minimally invasive interventions. The role of hybrid procedures, combining surgical and catheter-based techniques, is also being explored to minimize the invasiveness of treatment while ensuring effective relief of the obstruction. Furthermore, advances in prenatal imaging and fetal cardiology may enable earlier detection and potentially pave the way for fetal interventions in the future, although such approaches remain experimental.15

In conclusion, the management of Cor triatriatum has evolved considerably, with surgical resection being the mainstay of treatment for most patients. Minimally invasive approaches and medical therapies play adjunctive roles in the comprehensive management of this complex congenital heart defect. A multidisciplinary approach involving cardiologists, cardiothoracic surgeons, and intensivists is crucial for optimizing outcomes and ensuring long-term health in these patients.16

CONCLUSION

Cor triatriatum represents a rare but significant congenital cardiac anomaly characterized by the division of an atrium into two distinct chambers by a fibromuscular membrane, leading to variable degrees of blood flow obstruction. The pathophysiological consequences of this malformation are profound and depend on the extent of obstruction, the presence of associated congenital heart defects, and the patient's age at presentation. The clinical spectrum of Cor triatriatum ranges from asymptomatic individuals with incidental findings to neonates or infants presenting with life-

threatening heart failure or severe respiratory distress due to marked pulmonary venous hypertension.

Advances in imaging techniques have greatly enhanced our ability to diagnose and understand the complex anatomy of Cor triatriatum. Echocardiography, both transthoracic and transesophageal, remains the cornerstone for initial assessment, offering crucial insights into the presence and severity of the atrial membrane, as well as the overall cardiac structure and hemodynamics. Cardiac MRI and CT angiography provide further anatomical detail, especially in cases requiring intricate surgical planning or when associated congenital anomalies are present. While cardiac catheterization has become less common as a primary diagnostic tool, it retains a valuable role in hemodynamic assessment and in cases where non-invasive imaging is inconclusive.

Surgical intervention is the treatment of choice for symptomatic patients or those with significant hemodynamic compromise. Complete resection of the membrane and restoration of normal atrial anatomy are key to alleviating symptoms and preventing the progression of pulmonary hypertension and heart failure. Outcomes of surgical treatment are generally favorable, with low morbidity and high success rates, especially when performed in specialized congenital heart centers. The evolution of surgical techniques and the emphasis on early intervention have contributed to the improved prognosis observed in contemporary cohorts. However, long-term follow-up remains essential, as patients may develop complications such as atrial arrhythmias, and rare cases may require reintervention.

In certain cases, particularly in critically ill neonates or patients at high surgical risk, catheter-based interventions may provide a less invasive option to palliate symptoms or delay the need for surgery. Nevertheless, the efficacy and durability of these interventional approaches are variable, and more research is needed to establish their role in the definitive treatment of Cor triatriatum. Medical management, while not curative, is vital for stabilizing acutely decompensated patients and managing heart failure symptoms in preparation for definitive intervention.

The presence of associated congenital heart defects, such as atrial septal defects, persistent left superior vena cava, or anomalous pulmonary venous connections, adds complexity to the diagnosis and management of Cor triatriatum. A comprehensive, multidisciplinary approach is crucial, involving pediatric and adult congenital cardiologists, cardiothoracic surgeons, radiologists, and anesthesiologists to optimize outcomes and tailor treatment strategies to each patient's unique anatomy and clinical presentation.

Looking to the future, advancements in fetal imaging and prenatal diagnosis hold promise for earlier detection and potential in utero interventions, although these remain in the experimental stage. Research into the genetic and developmental mechanisms underlying Cor triatriatum may also yield insights that can improve diagnosis and treatment strategies. Additionally, further development of hybrid procedures combining surgical and interventional techniques may expand treatment options, particularly for complex or high-risk cases.

In conclusion, Cor triatriatum is a rare but clinically important congenital anomaly that demands a nuanced and highly individualized approach to management. While surgical correction remains the definitive treatment, ongoing advancements in imaging, interventional cardiology, and surgical techniques continue to improve outcomes for affected patients. A thorough understanding of the condition's pathophysiology, the implementation of a multidisciplinary care model, and vigilant long-term follow-up are essential to ensure that patients with Cor triatriatum lead healthy, productive lives. As our knowledge and technological capabilities advance, the management of Cor triatriatum will continue to evolve, offering hope for even better prognoses and quality of life for those affected by this complex congenital cardiac anomaly.

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