Case Report

A Rare Case of Pulmonary Artery Trunk Aneurysm

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Abstract

Background: Pulmonary artery trunk aneurysm (PATA) is a rare and complex vascular anomaly characterized by the abnormal dilation of the initial portion of the pulmonary artery, posing significant diagnostic and therapeutic challenges. **Purpose:** This clinical case report aims to describe the follow-up of a patient with PATA, emphasizing the role of imaging in diagnosis and monitoring, as well as discussing potential associations with other conditions. **Research Design:** The study is designed as a clinical case report, detailing the longitudinal follow-up of a single patient with PATA. **Study Sample:** The subject of this study is a 48-year-old female patient with a history of idiopathic hypertension who developed a PATA. **Data Collection and/or Analysis:** Since 2010, the patient underwent various imaging exams, including echocardiography, computed tomography, and catheter angiography, to detect and evaluate the aneurysm at different stages. **Results:** The imaging results indicated a progression of the aneurysm over time, underscoring the importance of imaging in the early identification and monitoring of PATA. The report also explores the possible association of PATA with conditions such as pulmonary hypertension, Behçet's disease, and Hughes-Stovin syndrome, highlighting the diagnostic complexity. **Conclusions:** Imaging diagnosis is crucial for the detection, characterization, and monitoring of PATA, providing essential information for selecting appropriate treatment options and achieving a satisfactory prognosis. An individualized treatment approach, considering both medical and surgical options, is necessary based on the clinical characteristics of each patient.

Keywords

pulmonary artery trunk aneurysm, pulmonary hypertension, chest angiotomography

Introduction

The pulmonary artery trunk aneurysm (PATA) is a rare and complex vascular anomaly involving the abnormal dilation of the initial portion of the pulmonary artery, a vital structure in the human cardiovascular system, resulting in significant hemodynamic alterations. The occurrence of arterial aneurysms is more common in other vascular areas such as the aorta, while pulmonary artery trunk aneurysms are less frequent and still pose a clinical challenge due to their complex nature and potential complications.[1,2]

Although it is an uncommon condition, PATAs have garnered increasing interest among the medical community due to the diagnostic and therapeutic challenges they pose. With the advancement of cardiovascular imaging and the growing awareness of this condition, there has been an increase in the identification and accurate diagnosis of these aneurysms. This, in turn, has led to a better understanding of their pathophysiology, clinical manifestations, and treatment options.[3]

Imaging diagnosis plays a fundamental role in detecting and evaluating PATAs. Through imaging tests, it is possible to determine the size, location, and extent of the aneurysm, assisting in

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risk stratification and planning appropriate treatment for each patient. There are various imaging modalities that can be used for this purpose, such as chest radiography, echocardiography, computed tomography angiography, and catheter angiography. The choice of imaging technique will depend on the case and resource availability; in some cases, a combination of different imaging modalities may be necessary to obtain a comprehensive assessment of PATAs.[2,4] Angiography is considered the gold standard for detecting pulmonary artery aneurysms. However, MR angiography is an excellent alternative because it is a safe, non-invasive test that can be used both for diagnosis and for monitoring pulmonary artery aneurysms.[5]

The aim of this study is to present a clinical case report on pulmonary artery trunk aneurysm, addressing the patient's symptoms, associated conditions, and the imaging diagnostic tests employed for the detection and characterization of the aneurysm. The case was approved by the Brazil National Research Ethics Committee (REC) "Plataforma Brasil", and the patient signed the Informed Consent Form (ICF) under number 6.679.884.

Incidence

The incidence of pulmonary artery aneurysms is difficult to determine accurately, as this condition is rarely described in the medical literature. It was first described by Bristowe in 1960, who identified a pulmonary artery aneurysm during an autopsy.[2] Today, the material published on the subject mostly consists of isolated case reports. The detection of pulmonary aneurysms is, in most cases, identified incidentally as a diagnostic finding. A study conducted in 1947 by Deterling and Claggett[6] indicated a prevalence of pulmonary aneurysms of less than 8 per 100 000 patients.

The average age of patients affected by pulmonary artery aneurysms is between 40 and 60 years. However, it is important to note that there is little robust material on the subject in the medical literature, which mostly consists of isolated case studies or those associated with other pathologies, not reflecting the actual incidence in the larger population.[2,7]

The etiology of pulmonary artery aneurysms is often related to pulmonary arterial hypertension, congenital heart diseases, and/or conditions that cause vascular disorders of the pulmonary artery, pulmonary hyperflow, post-stenotic dilation of the pulmonary valve, systemic arteritis, including Behçet's disease, trauma, and infections.[2,8]

The pulmonary artery is less prone to pathological dilations compared to the aorta because the blood pressure in the pulmonary artery is significantly lower than in the aorta. Since aneurysms often occur in vessels with high pressure, the low pressure in the pulmonary artery reduces the likelihood of aneurysm formation.[2,9]

The increased pressure in the pulmonary arteries in cases of severe pulmonary hypertension results in excessive stress on the walls of the blood vessels, progressively damaging their structure. This can lead to the dilation of the pulmonary artery, thereby increasing the potential for aneurysm formation.[2]

Case Report

Female patient, approximately 50 years old, who consumes alcohol regularly and is a former smoker, having quit in 2018 after 30 years of smoking. Diagnosed with idiopathic hypertension at the age of 20, she has been under outpatient follow-up at this institution since 2010. Despite extensive investigation, the etiology of the hypertension remains undefined.

At the first consultation in August 2010, the patient was referred for investigation of primary pulmonary hypertension. She reported that the diagnosis was made after experiencing syncope and tingling in the lower limbs (LL), with bulging of the pulmonary artery evident on chest X-rays and marked pulmonary hypertension with enlargement of the right chambers and trunk of the pulmonary artery on echocardiography. At that time, sildenafil and anticoagulation were prescribed, but she did not take the medications and missed the follow-up visit, continuing only with nifedipine.

The patient returned 8 years later (July 2018) with a history of precordial and left thoracic region pain, of strong intensity, starting 6 days before the consultation, associated with facial and left hemibody edema, as well as productive cough. A chest computed tomography was performed, which showed the presence of a fusiform aneurysm of the pulmonary artery. Through the exams conducted during hospitalization, the diagnosis of community-acquired pneumonia (CAP) was also made. Antibiotic therapy with amoxicillin-clavulanate, azithromycin, and sildenafil was administered. The patient was discharged 3 days later, in good general condition, eupneic, with an early outpatient follow-up, without a surgical proposal as per the cardiac surgery evaluation.

After almost 5 years (February 2023), the patient returned to the pneumology outpatient clinic with complaints of anxiety due to family stress, worsening dyspnea, and a history of orthopnea, polyuria, and nocturia. Considering the patient's history and worsening clinical condition, sildenafil, ambrisentan, and rivaroxaban medications were maintained, with the addition of spironolactone. Furthermore, a new battery of tests was requested to monitor the aneurysm and characterize the etiology of the disease, with primary suspicions for investigation including pulmonary hypertension, Behçet's disease, and Hughes-Stovin syndrome.

Next a list of exams and procedures performed since the beginning of her clinical follow-up in 2010 until the present moment are presented.

- Echocardiogram (2010): enlargement of right heart chambers, pulmonary hypertension, asymmetric pulmonary artery flow, and enlargement of the pulmonary artery trunk (6.7 cm).
- Catheterization (2011): Moderately elevated pulmonary artery pressure, decreased cardiac output, and increased resistance.

- Echocardiogram (2018): Left atrial and right heart chamber dilation, pulmonary hypertension, enlargement of the pulmonary artery trunk (8.8 cm).
- Chest computed tomography (2018): Pulmonary artery aneurysm with a maximum diameter of 9.4 cm extending from the trunk to its bifurcation, continuing as fusiform aneurysms of the right (up to 4.3 cm) and left (up to 3.4 cm) branches to their segmental branches. The right pulmonary artery and its main branches show discontinuous parietal calcifications, suggestive of long-standing pulmonary hypertension, along with minimal soft atheromatous plaques.
- Catheterization (2019): Moderate pulmonary hypertension with decreased cardiac output.
- Chest X-ray (2018): Shown in Figure 1, patient had increased cardiac silhouette, particularly in the left heart region.
- Echocardiogram (2020): Moderate pulmonary hypertension, right heart chamber dilation, grade II left ventricular diastolic dysfunction, concentric left ventricular hypertrophy, moderate pericardial effusion, and enlargement of the pulmonary artery trunk (8.4 cm).
- Chest computed tomography angiography (2023): Dilatation of the pulmonary artery trunk, with a caliber of up to 10.7 cm, and of the patent right (3.4 cm) and left (3.1 cm) pulmonary arteries. The trunk compresses the left bronchus, with signs of pulmonary hyperinflation on this side. Additionally, moderate dilation of the right heart chambers and moderate pericardial effusion are observed. Figure 2 depicts an axial cut of contrast-enhanced chest computed tomography at the level of the pulmonary trunk, while Figure 3 shows the three-dimensional reconstruction of the pulmonary artery trunk.
- No further action (2023-2024): Although an endovascular approach was indicated for the condition, the patient refused treatment. Instead, her pulmonary hypertension has been managed through outpatient follow-up and medical treatment.

Figure 4 presents the patients treatment algorithm. It involves monitoring symptoms, performing evaluations and tests, adjusting medications, and considering surgical intervention based on the severity and progression of pulmonary hypertension or aneurysm, with regular follow-ups to reassess the condition.

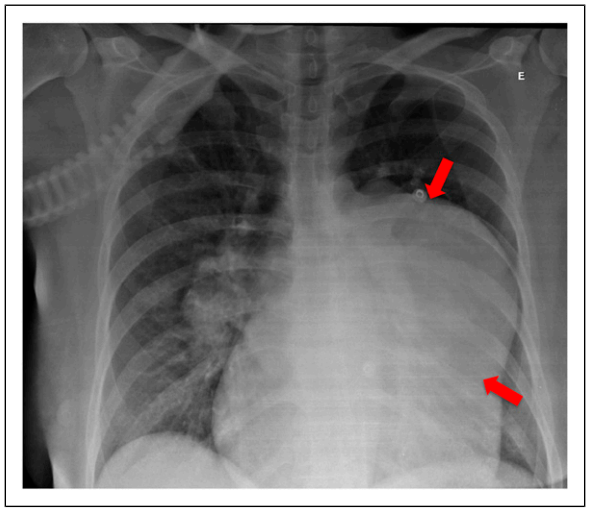

Figure 1. Chest X-ray demonstrating enlargement of the cardiac area, predominantly on the left side of the heart. (E denotes left). X-Ray performed in 2018.

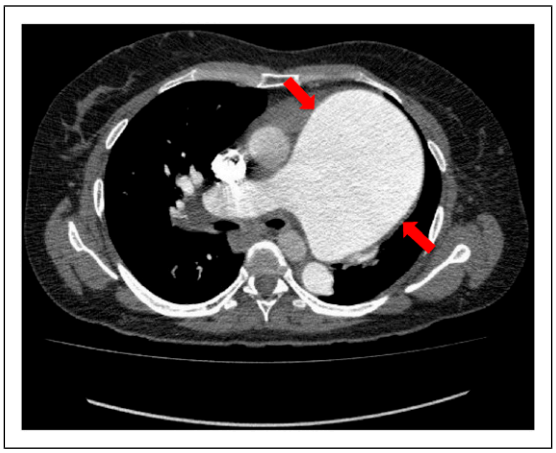

Figure 2. Axial section of contrast-enhanced computed tomography of the chest. The CT angiography of the chest revealed dilation of the pulmonary artery trunk, measuring up to 10.7 cm in diameter, compressing the left bronchus and showing signs of pulmonary hyperinflation on that side. CT performed in 2023.

Discussion

Although the exact etiology of PATA remains not entirely understood, it is believed that genetic and congenital factors play a significant role in its occurrence. Additionally, acquired conditions such as inflammatory and cardiovascular diseases may also contribute to the development of pulmonary artery trunk aneurysms.[2]

PATA is an abnormal and localized dilation of this artery, occurring due to weakness in the artery wall, which expands and becomes susceptible to rupture. Although PATA, pulmonary hypertension, Behçet's disease, and Hughes-Stovin syndrome are distinct diseases that can develop independently of each other, there are some situations where they may be related.[10,11]

Pulmonary hypertension is a condition that affects the arteries in the lungs, leading to an increase in blood pressure in this region. This occurs due to a series of changes in the walls

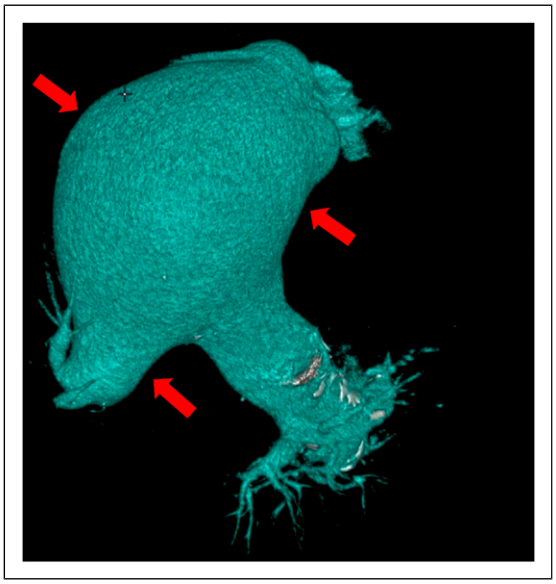

Figure 3. Three-dimensional reconstruction of the pulmonary artery trunk. The CT's 3D reconstruction reveals a pulmonary artery aneurysm with a maximum diameter extending from the trunk to its bifurcation, continuing as fusiform aneurysms on the right and left sides to their segmental branches. CT performed in 2023.

of the pulmonary blood vessels, resulting in narrowing or obstruction of blood flow. As a consequence, the heart needs to exert more force to pump blood through these restricted vessels, which can eventually lead to cardiovascular complications, including aneurysms, due to increased stress on the arterial walls.[12-14]

Behçet's disease is a rare but clinically significant condition, characterized as a chronic inflammatory disease that affects multiple systems. Although the classic symptoms of the disease mainly involve oral and genital mucosa, skin, and eyes, vascular involvement is a serious complication that can occur in up to 40% of patients. Systemic inflammation and activation of the immune system can lead to degeneration of the arterial wall and weakening of tissues, predisposing to the formation of aneurysms. Additionally, the aberrant immune response observed in Behçet's disease, with increased production of inflammatory cytokines and endothelial dysfunction, may contribute to the formation and progression of PATA.[5,15,16]

Hughes-Stovin syndrome, in turn, is characterized by the presence of recurrent deep venous thrombosis, mainly in the lower limbs, and the development of aneurysms in the pulmonary artery trunk. This is because this syndrome can manifest as a systemic vasculitis, in which chronic

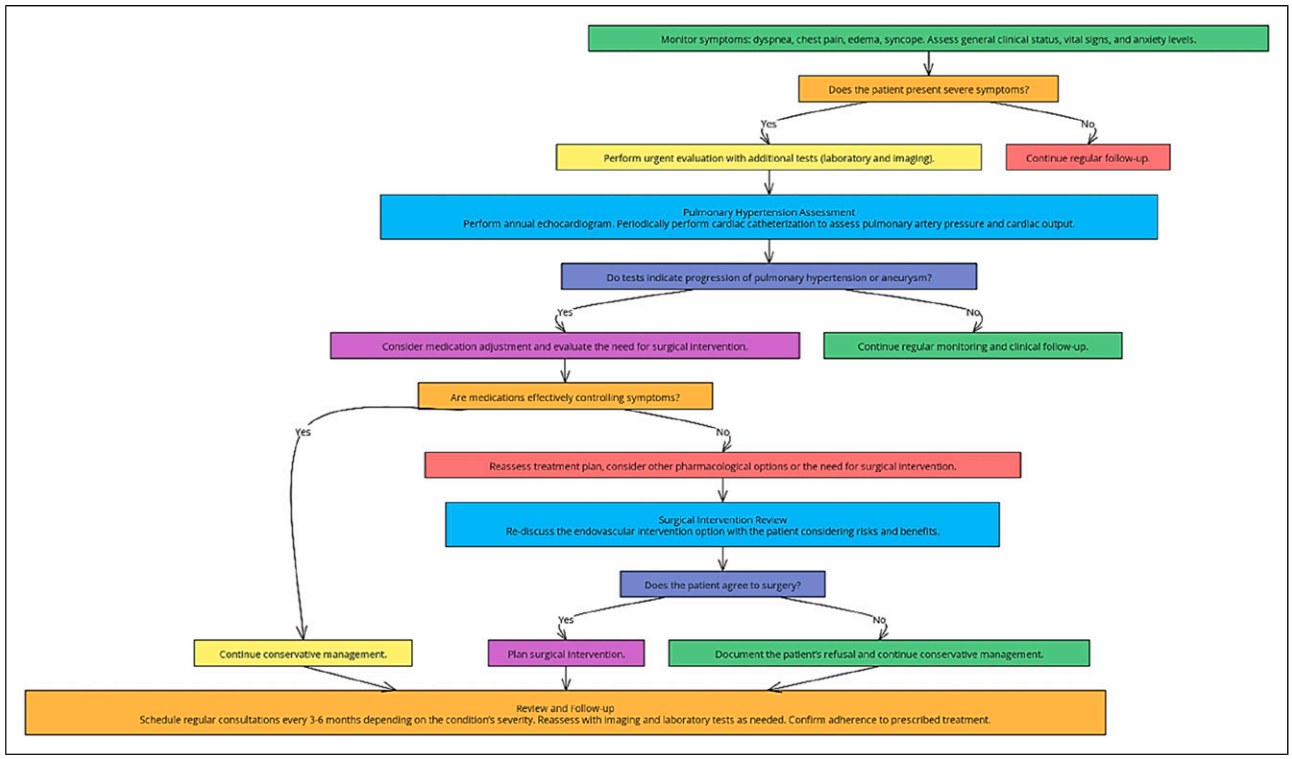

Figure 4. Patient's treatment algorithm.

inflammation affects the walls of blood vessels. This inflammation can lead to the formation of thrombi and subsequent aneurysmal dilation of the pulmonary arteries. Additionally, the presence of autoantibodies and immune dysfunction may be involved in the pathogenesis of Hughes-Stovin syndrome and in the formation of aneurysms.[17-19]

The clinical manifestation of PATA can vary widely, from asymptomatic to severe symptoms related to cardiovascular dysfunction. Common symptoms include dyspnea, fatigue, chest pain, and signs of right heart failure. Early diagnosis is crucial to prevent potentially fatal complications such as aneurysm rupture, heart failure, thrombosis, or pulmonary embolism. Imaging diagnosis involves the use of various radiological modalities to visualize and characterize the aneurysm.[2,11]

Chest radiography is usually the first imaging test performed to investigate pulmonary changes. Although not sufficient to confirm the definitive diagnosis of an aneurysm, chest radiography provides clues to PA trunk aneurysm, such as pulmonary artery dilation.[14] Echocardiography is also employed in the initial screening and follow-up of patients suspected of having PATA. It is an ultrasound technique that allows real-time visualization of cardiac structures, providing information about the size, shape, and function of the pulmonary artery trunk, including the detection of dilation or aneurysm.[11,20]

The treatment of pulmonary artery aneurysms remains controversial. Stable and asymptomatic patients are often monitored clinically for long periods, often more than 3 decades. The decision regarding the need for surgery remains debated, as there is still no consensus on the dilation diameter that would accurately indicate surgical intervention. Unlike aortic aneurysms, for which intervention is generally recommended when the diameter reaches or exceeds 60 mm, there is no established parameter for pulmonary artery aneurysms.[4]

Computed tomography is an imaging technique that uses X-rays to create detailed cross-sectional images of the chest. CT is an important tool in the diagnosis of PATA as it can provide precise information about the location, size, extent, and characteristics of the aneurysm. Additionally, CT can assist in the evaluation of associated complications, such as intraluminal thrombi, vessel obstruction, or compression of adjacent structures.[21-23]

Catheter angiography is an invasive procedure involving the insertion of a catheter into a peripheral artery, typically in the groin, which is guided to the pulmonary artery trunk. Contrast medium is injected to visualize the pulmonary arteries and identify the aneurysm. The technique allows for detailed imaging and the possibility of performing therapeutic procedures simultaneously.[11,20,24]

The treatment of PATA is complex and should be individualized according to the clinical characteristics of each patient. Therapeutic options include drug therapy to control symptoms and complications, as well as surgical interventions such as open surgical repair or less invasive endovascular techniques. The choice of treatment depends on various factors such as the size of the aneurysm, the presence of symptoms, cardiac function, and patient preference.[25,26]

Drug therapy may be recommended to treat symptoms associated with PATA, such as dyspnea or pulmonary hypertension. Medications may include bronchodilators, diuretics, anticoagulants, or drugs to control elevated pulmonary arterial pressure. Drug therapy can be used as a complementary approach to clinical management or in conjunction with other therapeutic modalities. In specific cases, surgery may be indicated to treat PATA. Surgery may involve resection of the aneurysm and reconstruction of the pulmonary artery trunk with a vascular graft. This procedure aims to eliminate the aneurysm and restore normal artery function. However, surgery is considered a more invasive option and may be associated with risks and complications.[27,28]

Final Considerations

The dilations of the pulmonary artery, characterized as aneurysms, although less frequent than in other vessels, have a great potential to generate complications, making it difficult to choose the appropriate treatment and consequently achieve a satisfactory prognosis for the patient.

As it is an uncommon condition, we can observe through the reported case that the diagnosis of the aneurysm was constructed as the patient presented at this institution over the cited time interval (2010-2023), with the aid of the combination of different imaging diagnostic techniques, from a simple chest radiography to more sophisticated modalities such as angiography. This allowed for the detection and monitoring of the evolution of the aneurysm. In light of the proposed case report, we can infer that imaging diagnosis was present throughout the investigation of the pathology and patient monitoring, proving to be essential in the detection and evaluation of the pulmonary artery trunk aneurysm, as well as in assisting in the choice of therapeutic approach that best suits these patients.

Regarding the treatment of the pulmonary artery trunk aneurysm in the reported patient, although an endovascular approach was indicated for the condition, the patient chose not to follow this treatment. Instead, the management of her pulmonary hypertension has been conducted through outpatient follow-up and medical treatment.

Currently, the patient's treatment plan includes the use of the following medications for controlling pulmonary hypertension: Sildenafil, a specific vasodilator for pulmonary hypertension; Ambrisentan, an endothelin receptor antagonist; Rivaroxaban, an oral anticoagulant; Spironolactone, a diuretic that helps control fluid volume and blood pressure.

At the moment, there is no proposed plan for surgical intervention, considering the patient's refusal to undergo the endovascular procedure, thus she continues with outpatient

follow-up with treatment adjustments as necessary to manage pulmonary hypertension and its associated complications.

Learning Outcomes

Our article aimed to provide important and novel learning outcomes regarding pulmonary artery trunk aneurysms. Specifically, it focuses on delivering a comprehensive understanding of diagnostic approaches and imaging modalities essential for evaluating PATAs, fostering proficiency in interpreting radiological findings across various modalities such as radiographs, echocardiograms, computed tomography scans, and angiography. Moreover, it enhances knowledge regarding the clinical manifestations, differential diagnoses, and management strategies associated with PATAs, emphasizing the interdisciplinary collaboration required for optimal patient care. Furthermore, the article underscores the significance of recognizing potential complications and prognostic factors influencing treatment outcomes, contributing to a deeper understanding of this rare but clinically significant vascular anomaly.

Declaration of Conflicting Interests

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Supplemental Material

Supplemental material for this article is available online.

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