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Successful Surgical Repair of Pulmonary Artery Sling in a 10-Month-Old Infant: A Case Report

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Abstract

Pulmonary artery sling (PAS) is a rare congenital vascular anomaly in which the left pulmonary artery originates from the right pulmonary artery, often causing airway compression and respiratory symptoms in infancy. We report a 10-month-old infant presenting with respiratory symptoms since early infancy. Transthoracic echocardiography and computed tomography angiography confirmed the diagnosis of pulmonary artery sling. Surgical repair with reimplantation of the left pulmonary artery was successfully performed. The postoperative course was uneventful, and the patient was discharged without complications. This case underscores the importance of early diagnosis using appropriate imaging modalities, careful surgical planning, and multidisciplinary management to achieve favorable outcomes in infants with pulmonary artery sling.

Keywords: Pulmonary Artery Sling; Stridor; Surgical Repair; Vascular rings; Congenital Vascular Anomalies; Tracheal Compression

Introduction

Pulmonary artery sling (PAS) is a rare vascular anomaly characterized by the abnormal origin of the left pulmonary artery from the right pulmonary artery, instead of the pulmonary trunk [1,2]. Left pulmonary artery runs posteriorly between the esophagus and trachea, causing a series of compression-related symptoms. Because these symptoms are often atypical, PAS usually remains underdiagnosed or frequently misdiagnosed as a respiratory pathology [3]. In this article, we report a case of diagnosis and successful surgical treatment of pulmonary artery sling in a 10-month-old infant who presented with dyspnea and stridor.

Case presentation

A 10-month-old boy was admitted to our hospital for with a history of cough, shortness of breath, recurrent stridor, that worsens during physical exertion. These symptoms appeared at 6 months of age. The patient had no remarkable prenatal history and was born at 39 weeks of gestation and a birth weight of 3,840 g. His past medical history determined that he had been hospitalized to outside hospital 2 times during the last year due to respiratory distress.

Physical examination revealed subcostal retraction, nasal flaring, and no heart murmur. Chest radiograph were unremarkable. Complete blood count (CBC) was normal. Transthoracic echocardiography demonstrated that the left pulmonary artery (LPA) was originating from a right pulmonary artery (RPA) (**Figure 1**). CT confirmed PAS (**Figure 2**). The left pulmonary artery arose from the posterior aspect of the right pulmonary artery then crossed the midline between the trachea and oesophagus to reach the left pulmonary hilum. Transthoracic echocardiography and CT scan revealed no other cardiac abnormalities. Bronchoscopy was not performed.

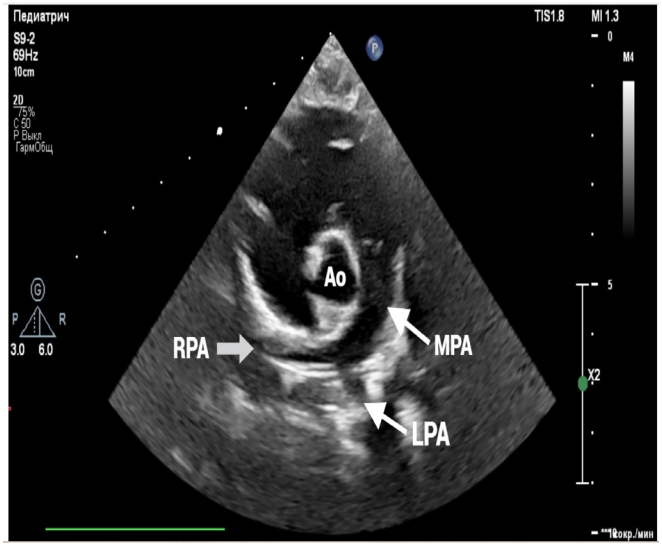

Figure 1: Echocardiography showing the abnormal origin of LPA from RPA. MPA, main pulmonary artery; LPA, left pulmonary artery; RPA, right pulmonary artery; AO, aorta.

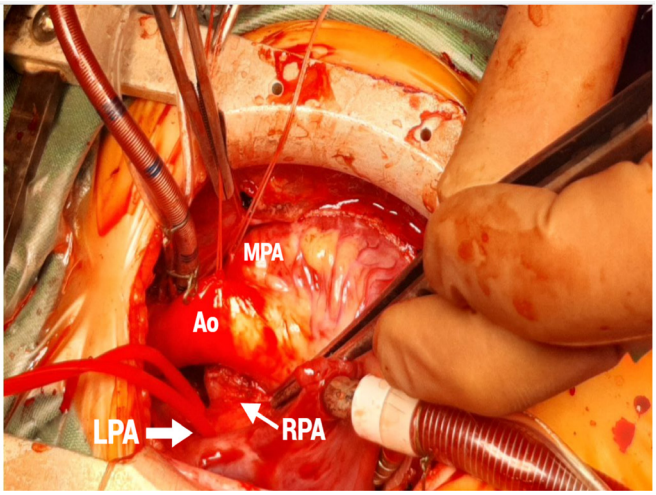

Figure 3: Intraoperative photo showing abnormal arising of the LPA from the RPA. MPA, main pulmonary artery; LPA, left pulmonary artery; RPA, right pulmonary artery; AO, aorta.

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After comprehensive preoperative assessment the patient underwent surgical correction of PAS. A median sternotomy was performed. After heparin administration and canulation of aorta and right atria cardiopulmonary bypass was initiated. Visually, the left pulmonary artery emerged from the posterior surface of the right pulmonary artery behind the superior vena cava and then passed behind the trachea (**Figure 3**). Without cardiac arrest under normothermia and on a beating heart, the LPA was detached from the RPA. Incision of the right pulmonary artery was sutured completely. LPA was dissected free from surrounding tissues and removed from under the trachea. Incision was made in the left side of the main pulmonary artery and LPA was repositioned to the main pulmonary artery in the proper anatomical location (**Figure 4**). The patient was weaned from CPB without complication. Finally, heparin neutralization, hemostasis, and chest closure were performed. The patient was extubated 4 hours after surgery and transferred from the ICU the next day. After surgery, all respiratory symptoms were eliminated and the patient was discharged ten days after surgery in a good condition. Postoperative CT (**Figure 5**) confirmed good geometry of the pulmonary artery branches and patency of the anastomosis.

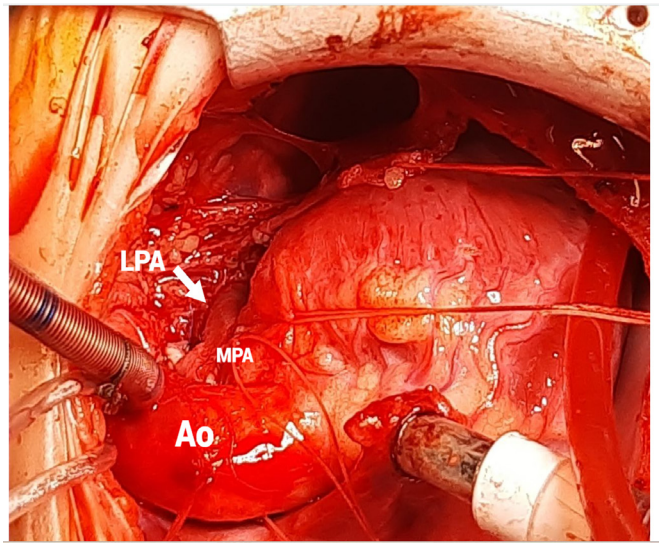

Figure 4: Intraoperative photo showing position LPA after reimplantation. MPA, main pulmonary artery; LPA, left pulmonary artery; AO, aorta.

Discussion

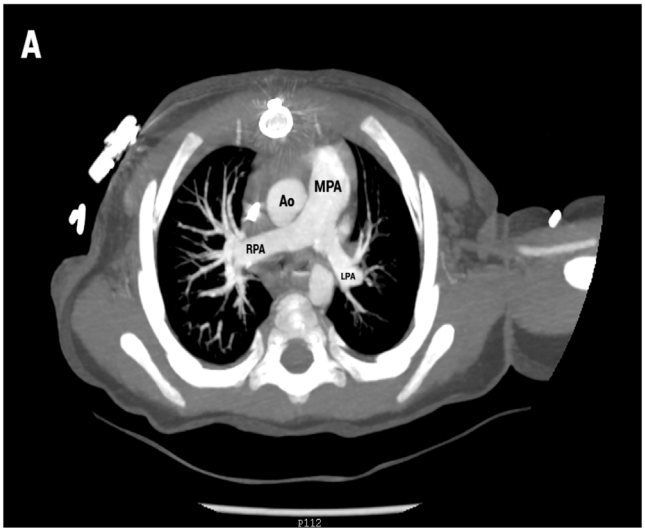

Figure 5: Post-operative CT angiography. (A) Axial image shows of the left pulmonary artery arising from the main pulmonary artery.

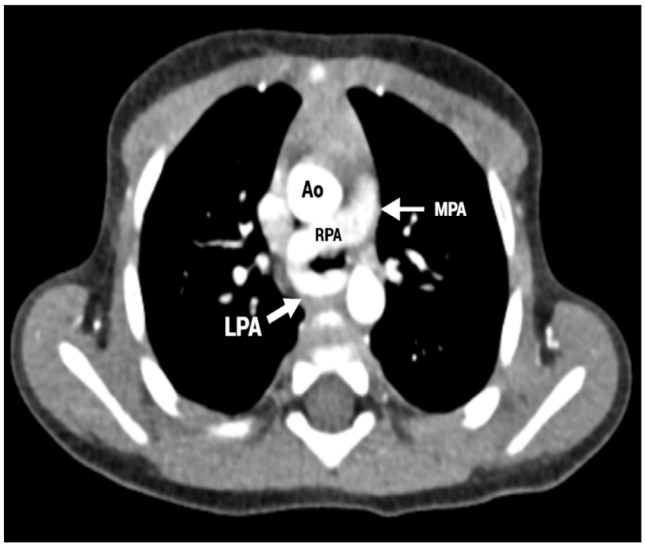

Figure 2: Pre-operative computed tomographic angiography (CTA) showing anomalous origin of the LPA from the posterior aspect of the RPA. The aberrant LPA runs behind the trachea as it courses to the left pulmonary hilum. MPA, main pulmonary artery; LPA, left pulmonary artery; RPA, right pulmonary artery; Ao, ascending aorta.

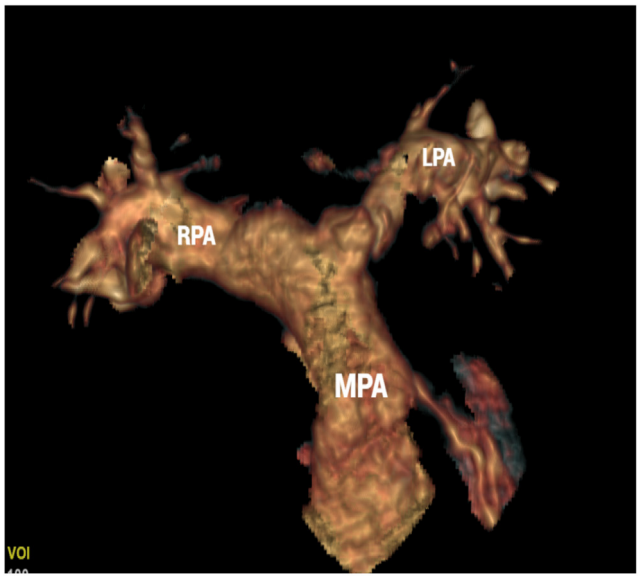

Figure 5: Post-operative CT angiography. (B) 3D image. MPA, main pulmonary artery; LPA, left pulmonary artery; AO, aorta.

Pulmonary artery sling is a rare vascular anomaly that occurs with a frequency of 59 per million live births, with 90% of cases diagnosed in the first year of life [4]. The anomaly is characterized by the arising of the left pulmonary artery from the right pulmonary artery, passing between the trachea and esophagus. The main pathophysiological mechanism for the appearance of symptoms is compression of surrounding structures. Glaevecke and Doehle first described the anomalous origin of the left pulmonary artery from the right pulmonary artery in 1897 [5]. Dr. Willis Potts performed the first successful surgical repair of PAS on a 5-month old through a right thoracotomy approach in 1953 [6].

The anomaly may be asymptomatic or manifest with a number of nonspecific symptoms associated with compression of the esophagus and trachea, and therefore the accurate diagnosis is often established in older children. Cases of diagnosis of the anomaly in adulthood have been described [7]. PAS may occurs as isolated anomaly or may be associated with some congenital heart defects (atrial or ventricular septal defects, aortic arch anomalies) and tracheobronchial defects (complete tracheal rings, bridging bronchus) [8,9].

Echocardiography allows visualization of the anomalous origin of the left pulmonary artery, but is not the main method for diagnosing PAS. The main imaging method for PAS is computed tomography (CT) with contrast and magnetic resonance imaging (MRI), which allow for a detailed assessment of the anatomy of the defect and the degree of tracheal compression. Another method used to assess the degree of tracheal compression is bronchoscopy. Bronchoscopy was not used in our clinical case.

PAS has a high mortality rate without surgical intervention, especially in newborns. In our opinion, PAS is an absolute indication for surgical repair, even in the absence of symptoms. Performing surgical intervention at an early age avoids secondary changes in the tracheobronchial tree, namely tracheo- and bronchomalacia, which significantly worsen long-term results.

The goal of surgical treatment is to reimplant the left pulmonary artery into the correct anatomical position in the pulmonary artery trunk. Traditionally, the operation is performed under cardiopulmonary bypass and on a beating heart. Cases of surgical treatment of PAS without cardiopulmonary bypass have been described [10].

In our clinical case, we present a case of diagnosis and successful surgical treatment of a 10-month-old infant who was treated twice a year in another medical institution for respiratory distress. High clinical suspicion of cardiologists from another medical institution allowed to establish a diagnosis at an early age and perform surgical treatment in a timely manner.

Conclusion

Pulmonary artery sling is a serious congenital anomaly that requires early diagnosis and surgical intervention to prevent secondary changes in the tracheobronchial tree. Thanks to modern diagnostic and surgical methods, most children after surgery have a favorable prognosis. However, to ensure long-term success, it is necessary to carefully monitor patients in the postoperative period, timely detecting and correcting possible complications.

Patient Consent

Written informed consent was obtained from the patient for publication of this case and accompanying images.

Conflicts of Interest

The authors declare no conflict of interest.

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