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CASE REPORT

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Successful transcatheter treatment of large right pulmonary artery to left atrial fistula: a case series and literature review

Hojjat Mortezaeian[1], Farshad Jafari[1], Mahmoud Meraji[1], Avisa Tabib[2], Hamid Reza Pouraliakbar[1] and Fatemeh Naderi[3*]

Abstract

Introduction Cyanotic congenital heart diseases are among the most serious anomalies among newborns. A rare type of this condition is direct communication between the right pulmonary artery and left atrium, which presents mostly in adolescence and adulthood. Large shunts, however, should be corrected as soon as possible, considering their potential to cause congestive heart failure.

Case Presentation Two 2- and 13-year-old patients with this pathologic communication complained of exertional dyspnea and central cyanosis. Their physical exams were prominent, with a low oxygen saturation level. The diagnostic modalities used, electrocardiogram, chest X-ray, echocardiogram, CT scan, and selective angiography of the pulmonary arteries, showed right-to-left abnormal blood flow through the right pulmonary artery and left atrium shunt. Finally, both patients were treated successfully by a transcatheter occluder without any complications or follow-up complaints.

Discussion The right pulmonary artery and left atrium abnormal congenital connections are rare causes of central cyanosis, mostly present with exertional dyspnea and cyanosis during adolescence or early adulthood. Transthoracic echocardiography, contrast-enhanced CT scans, and angiography of the pulmonary arteries make the diagnosis. The treatment has emerged during the last two decades, shifting from surgical treatment for severe cases to interventional percutaneous strategies, leaving the surgery for cases with no appropriate location for the application of interventional therapy.

Conclusion Considering the potentially life-threatening complications of the untreated right pulmonary artery and left atrium fistulas, such as thromboembolic events, early diagnosis is crucial. However, treating these patients is not always straightforward, and strategies should be selected based on the connecting fistula's anatomical features, location, size, and tortuosity. Although endovascular treatment is the preferred option, some patients need to be treated with surgical procedures due to their structural characteristics.

Keywords Interventional Cardiology, Congenital heart disease, Vascular shunt, Case report

*Correspondence:
Fatemeh Naderi
fatemeh_naderi2010@yahoo.com
[1]Rajaie Cardiovascular Medical & Research Center, Iran University of Medical Sciences, Tehran, Iran
[2]Heart Valve Disease Research Center, Rajaie Cardiovascular Medical and Research Center, Iran University of Medical Sciences, Tehran, Iran
[3]Rajaei Cardiovascular Medical and Research Center, School of Medicine, Iran University of Medical Sciences, Tehran, Iran

Introduction

Congenital heart disease (CHD), defined as an abnormal structure of the heart or intrathoracic great vessels, is the leading cause of death caused by congenital malformations in childhood [1]. These pathologic abnormalities are categorized as either (1) non-cyanotic or (2) cyanotic congenital heart disease (CCHD). The pathophysiology of CCHD is the mixture of deoxygenated pulmonic blood flow with systemic blood; however, the etiology of CCHD has not been identified thoroughly [2]. Except in 15–20% of cases, which are mainly due to trisomy of 13,18,21, and Turner syndrome, other cases have been associated with environmental factors such as the mother's exposure to toxins, viral infections, or other diseases during or before pregnancy [1]. Almost 0.2–0.3% of live births are affected by CCHD cases, which are mostly one of these five subtypes of Tetralogy of Fallot (consisting of 7–10 of all CHDs), Transposition of the Great Arteries (accounting for 5–7% of CHDs), Truncus Arteriosus (seen in less than 1% of CHDs but 4% of critical ones), Tricuspid Atresia (0.3–0.5% of CHDs), and Total Anomalous Pulmonary Venous Return. Among these pathologies (rarely reported) [2]. However, since most cases of TOF are diagnosed a few months after birth, the most commonly seen CCHD in neonates are TGA patients [3]. One of the rare causes of CCHD is the direct communication of the left atrium and right pulmonary artery (RPA-LA) by an anomalous artery, which mainly originates from the posterior part of the PV and is connected to the LA. The clinical presentation of this disease is mainly observed in adolescence since the flow is not prominent until that age. On the other hand, a more significant shunt can lead to the emergence of symptoms in younger ages and indicate emergent corrective surgery [4, 5].

This study presents two rarely reported cases of RPA-LA diagnosed in the age groups mentioned above, along with their presentation, diagnosis, and therapeutic procedures.

Case presentations

Case 1

Patient's history and physical examination A 32-month-old male was brought to the emergency department of the tertiary congenital cardiac center due to central cyanosis. His cyanosis had been diagnosed at birth. However, since there were negligible, no follow-up had been performed by his family. His parents described cyanosis as a progressive process that was more prominent in the central parts of the body. No remarkable past medical or familial history or exposure to toxins and drugs was mentioned by the patient's parents. In his physical exam, he was 13 kg (Kg), height of 95 cm, and his blood oxygen saturation was 80% in room air. The patient was tachypneic (RR:28/per minute), and other vital signs were within the normal range (PR:95 per minute and T:37). There were no other remarkable abnormal findings in his physical exam.

Methods

In the initial laboratory findings, elevated Hemoglobin of 18.7 gr/dL (Normal range: 12–16), hematocrit (53.4%), marginally increased Lymphocytosis of 51% (Normal range: 20–45%), and low MCHC of 28.6 (Normal range: 32–36%) were detected. No other remarkable pathologic finding could be seen. The electrocardiogram was normal. The poster-anterior view chest X-ray of the patients showed double shadow, cardiomegaly, and decreased pulmonary vascular marking (PVM) (Fig. 1.A). Due to his cyanotic symptoms and low oxygen saturation, the patient underwent transthoracic echocardiography (TTE) to detect a potential structural abnormality. Although no intracardiac defects were detected, investigations revealed a dilated right pulmonary artery (RPA) larger than the left pulmonary artery (LPA). The ejection fraction (EF) was 60%, and a large orifice in the posterolateral aspect of LA with a 15 mm diameter was observed. Additionally, the size of the RPA was measured to be 20 mm. In contrast, the LPA diameter was estimated to be 8 mm (Table 1). Due to the dilation of the RPA, the possibility of a pulmonary arteriovenous (AV) fistula was raised, and a contrast TTE (CTTE) was performed (Fig. 1.B), which showed a delayed filling of the left heart and an extracardiac shunt (Table 1.C).

Therefore, a contrast-enhanced computed tomography angiography (CECTA) was performed to determine the shunt's detailed anatomy and measure the defect's size of RPA. Confluent MPA (16 mm) was detected in the CTA. Moreover, the ostium of dilated RPA was measured at 17.5 mm and its hilum at 16 mm, with the RPA connected to an aneurysmal right inferior pulmonary vein (RIPV) via the neck about 8 mm. On the other hand, the ostium of the LPA was 8 mm, and its hilum was 9.5 mm. It was also noted that all four pulmonary veins are connected to the left atrium.

Finally, the medical team decided to put the patient on a list of urgent endovascular procedures as a candidate for transcatheter intervention to impede RPA-LA communication. The AMPLATZER™ Septal Occluder device (ASO; Abbott/ St. Jude, St. Paul, MN, USA) size 13 mm and a 7 French sheath were used for the procedure. After the intervention, the patient's oxygen saturation increased immediately (80–92%). RPA injection in lateral view showed a good device position (Fig. 1.D).

Outcomes and Follow-Up The day After the completion of the procedure, the CTTE was repeated, revealing a delayed negligible right-to-left flow due to a small residual shunt around Amplatzer. The patient was discharged two days after the procedure. The 4-month follow-up outpa-

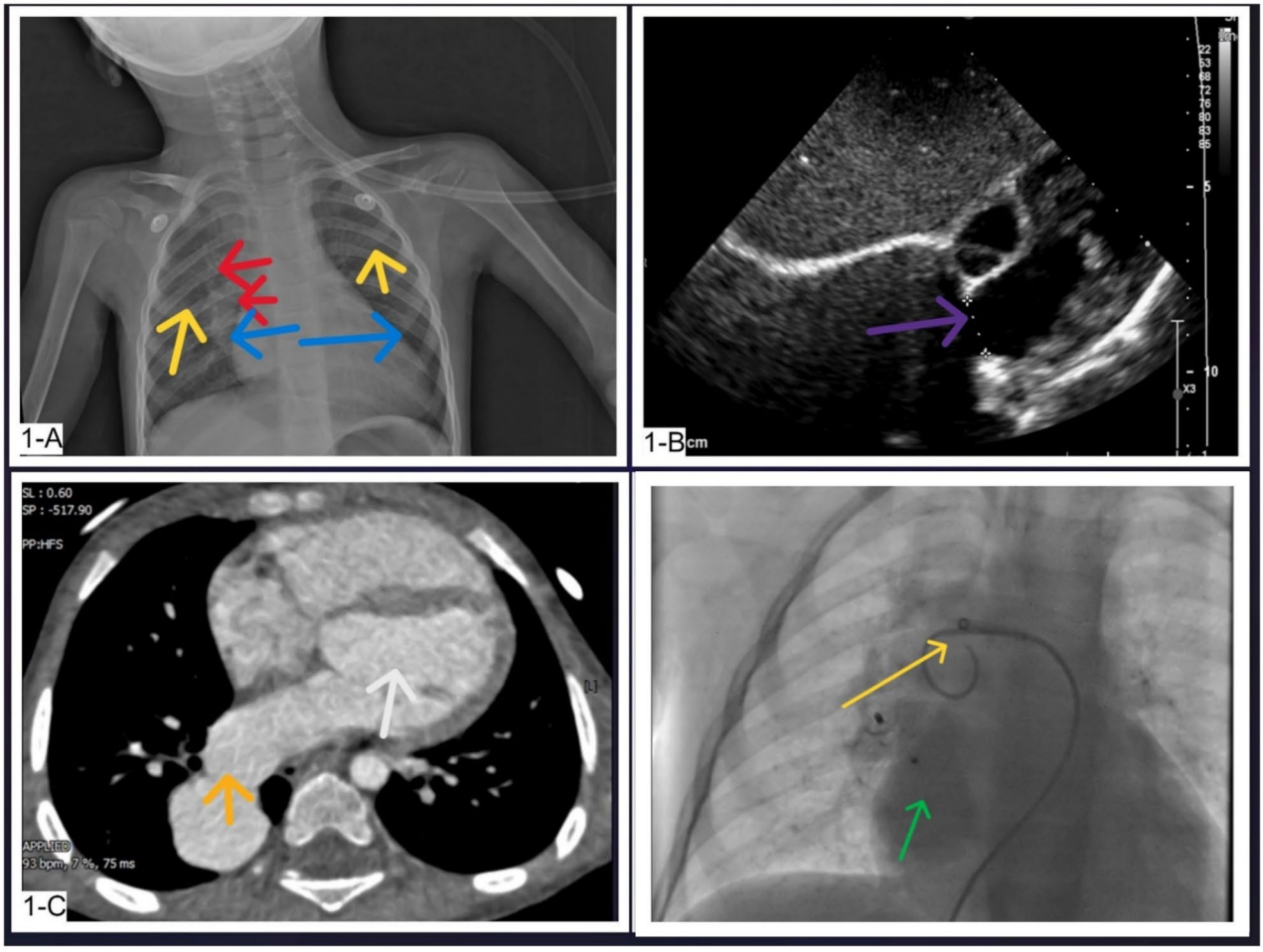

Fig. 1 (**A**) Patient's Chest X-ray showed double shadow (Red arrows), cardiomegaly (Blue arrows), and decreased pulmonary vascular marking (Yellow arrows), (**B**) echocardiogram showed the fistular between RPA and LA (purple arrow), (**C**) CT angiography of the chest shows dilated RPA (orange arrow) and the attachment of all (four) pulmonary veins to the left atrium (white arrow) (Tract entrance), (**D**) Release of the device in endovascular closure of the right pulmonary artery (yellow arrow) to the left atrium (green arrow) shunt and the closure of the shunt

Table 1 TTE findings at his first presentation to the hospital

Variable	Evaluation
Positions, situs, Ventricular looping	Levocardia, situs solitus, and D-ventricular looping
SVC and IVC Size	Normal
Pulmonary veins	Three pulmonary veins with a normal return to the LA
LA and LV size	Normal
LVEF	60%
RA and RV	Mild enlargement with normal function
AV valves	No MR/Mild TR PPG: 25mmHg
Great arteries and semilunar valves	NRGA, no AI or AS, Mild PI, PPG:20mmHg
Left Pulmonary artery	8 mm
Right Pulmonary artery	20 mm (dilated)
Septum	PFO: left to right shunt, No VSD
Shunt	No PDA
Aortic Arch	Left arch, No COA
Recommendation	presence of cyanosis◊contrast echo

tient evaluation demonstrated his condition improved dramatically. The oxygen saturation was 98%, and no new symptoms or physical examination findings were detected. A control TTE demonstrated no residual right-to-left, the fistula was occluded entirely, and no newly emerging abnormality or structural defect was detected.

Case 2

Patient's history and physical examination A 13-year-old male was brought to the emergency department of a tertiary cardiology hospital, complaining of dyspnea and palpitation, more prominently felt on exertion. The mild symptoms had begun several months before his presentation. However, the symptoms had been progressive, and the patient had difficulty doing his daily routine activities and could not participate in the physical exercises at his school during the past several weeks. No remarkable past medical or familial history or exposure to toxins

and drugs was mentioned by the patient's parents. In the patient's physical exam, central cyanosis (more prominent around lips) was seen in the evaluation of his general appearance. Assessment of the vital signs revealed an oxygen saturation of 65% in room air. The clubbing of the fingers and toes could be seen, but his other symptoms were unremarkable.

Methods His ECG showed normal sinus rhythm. In the evaluation of laboratory data, the only remarkably out-of-range serum markers were the hemoglobin level of 22 g/dL (Normal range: 12–16), and his PA CXR showed an increased cardiothoracic ratio (CTR) (Fig. 2.A). His TTE at this presentation showed the three pulmonary veins with a normal return to LA, Mild TR, and mild RAE and LAE. His holter monitoring showed an average heartbeat of 90 (Min:53 and Max: 141) and an SDNN of 123. Baseline ECG showed sinus rhythm with wide QRS. Infrequently, premature ventricular contracture (PVC) (Single and couplet) and premature atrial contracture (PAC) were seen. His cardiac exercise test showed no abnormal findings except PVCs at rest and recovery. Due to continued cyanosis, dyspnea, and palpitation, severe polycythemia (Hb:22 mg/dL, HCT:72%, RBC; $8.3*10^6$/μl), the TTE was repeated, which revealed a larger RPA than the LPA, suggestive of an abnormal extracardiac shunt. Mild LAE and a large orifice (18 mm) were observed in the posterior aspect of the LA. The CTTE demonstrated bubbles in the LA within three cardiac cycles and opacification of RA, which was considered a sign of structural abnormality. Furthermore, CTA of the pulmonary arteries (PACTA) revealed the ostium of the dilated RPA was 20 mm, and its hilum was 18 mm, with the RPA connected to an aneurysmal right inferior pulmonary vein (RIPV) via the neck approximately 12 mm. The ostium of the left pulmonary artery was 11 mm, and its hilum was 13 mm. It was also stated that all four PVs were connected to the LA. The Cardiac CTA demonstrated SDS and LV hyperaberculation. A PAVF type IV and RPA to LA fistula was detected.

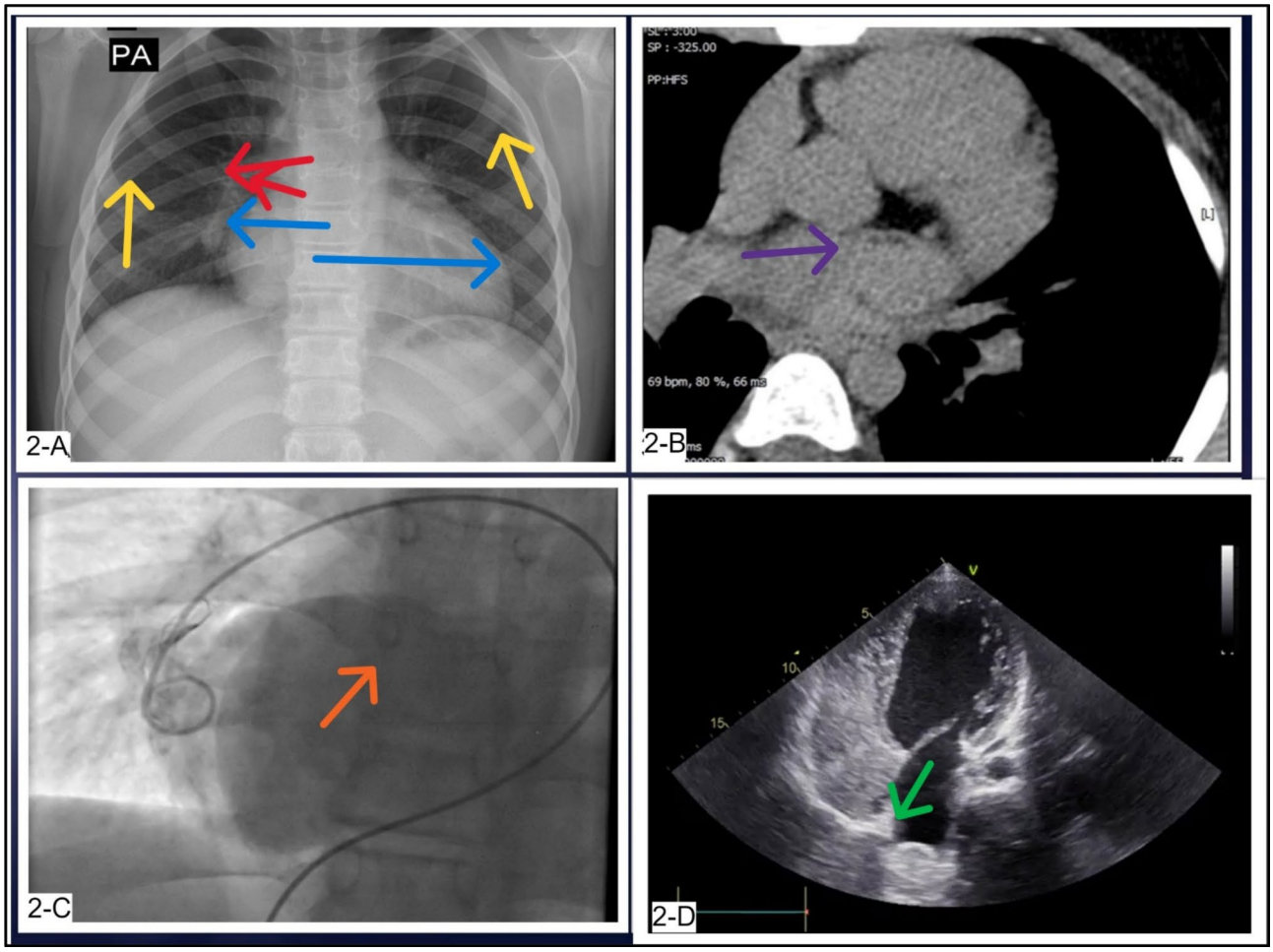

Fig. 2 (**A**) Chest X-ray shows double shadow (Red arrows), cardiomegaly (Blue arrows), and decreased pulmonary vascular marking (Yellow arrows), (**B**) Pulmonary CT angiography shows the fistula between the left atrium and the right pulmonary artery (purple arrow), (**C**) Pre-procedure angiography confirmed the diagnosis of RPA-LA fistula (orange arrow), (**D**) Post-procedure contrast echocardiographic shows good device position (green arrow)

Reduced LV function, Mild MR, and mild TR were other findings of the CTA (Fig. 2.B).

Considering all signs, symptoms, and diagnostic modalities' outcomes, the patient was determined to be a candidate for percutaneous intervention to impede the RPA to LA shunt. In the catheterization laboratory, local anesthesia was done, and femoral arterial access was obtained. The angiography of the PAa confirmed the diagnosis of RPA-LA fistula (Fig. 2.C). Using an Amplatzer Septal Occluder device (LifeTech Scientific Co., Shenzhen, China) size 20 mm and sheet 10 French, the connection between RPA to LA was occluded without any complication. The contrast TTE evaluation was performed after the procedure to control the appropriateness of the structures, which showed a good device position (Fig. 2.D). Moreover, considering the patient's continuous arrhythmia, the patient underwent an ablation of the abnormal foci, which was successful, and abnormal PVCs were not observed after the procedure.

Outcomes and Follow-Up The patient's oxygen saturation improved immediately after the procedure, from 65 to 95%. In the outpatient follow-up visit, the clinical condition was declared to have improved significantly, and the exertional cyanosis and dyspnea disappeared after the occlusion of the fistula. In addition, the control CTTE showed no residual shunt.

Discussion

This study presents two cases of RPA-LA fistula, which were approached successfully by the AMPLATZER™ Septal Occluder device (in the first case) and Amplatzer Septal Occluder device (LifeTech Scientific Co., Shenzhen, China) size 20 mm. The noteworthy aspect of the first case reported in this study is that it is one of the first RPA_LA cases reported to occlude the connection through interventional procedures due to the patient's young age. Both patients' treatment and follow-up courses were uneventful, and they both have recovered well and experienced significant clinical improvement. Cyanotic congenital heart diseases (CCHD) are one the most fatal types of congenital abnormalities during early childhood due to their relatively high rate of morbidity and mortality and the complexity of their management [6]. One rarely-reported type of CHD is the direct communication between the RPA and the LA (RPA-LA Shunt). Four types of RPA-LA are identified based on their anatomical features and the presence or absence of structural abnormalities. The anatomical features [7].

Patients with RPA-LA fistula more commonly present with signs and symptoms such as central cyanosis, clubbing of fingers, exertional dyspnea, silent precordium, normally second heart sound, nonspecific murmur on the left or right axillary area, an abnormal roentgenographic density in the right or left pulmonary hilum [5, 8]. The prevalence of these presenting signs and symptoms might differ considering the age. Among adolescents and early adulthood, complaining of central cyanosis with clubbing of fingers and exertional dyspnea are the most common presentations of this anomaly [9]. The emergence of these pathologic symptoms is attributed to the right to the left shunt of the deoxygenated blood to the systemic circulation and inducing the mentioned presentations due to the high hemoglobin level caused by the lack of oxygen supply to the organs. The reported range of oxygen saturation of the blood of these patients is mostly between 65 and 82% [8]. Other presentations such as neurologic sequels due to thromboembolic events (in 20% of patients), congestive cardiac failure, infective endocarditis, non-neurologic thrombo-embolic episodes, and even sudden hemodynamic instability and death due to aneurysmal dilatation of the fistula and rupture [10].

Diagnosis of this condition is not always straightforward and requires multimodality diagnostic measures. In laboratory findings, polycythemia and low arterial oxygen saturation are usually seen [8]. ECG and CXR can give hints about the diagnosis but cannot confirm it. In previous studies, various changes of CXR have been reported; however, a bulge beyond the right heart border is usually seen with RPA-LA fistula [9]. Mostly, the suspiciousness of any congenital or traumatic structural heart detection requires the evaluation with TTE as an accessible guiding tool [11]. The pitfall mentioned for the TTE to diagnose this structural abnormality is that it is operator-dependent and not as specific as the CT scan [9]. The bubble echo can also be useful in identifying the fistula between the RPA and LA [12]. Agitated saline echocardiography has been evaluated as an inexpensive and available diagnostic tool for diagnosing intracardiac and extracardiac fistula. Previous studies have approved its safety and advantages, such as the high sensitivity and differentiation of different concomitant anomalies. The point of this method is the necessity of performing provocative maneuvers that increase right atrial over left atrial pressure [13].

On the other hand, confirming the presence of RPA-LA fistula needs more accurate diagnostic tools such as CTA of pulmonary arteries. CTA normally shows the filling of LA in the early pulmonary phase and the observation of the fistula [14]. Zhu J. et al., in a study on this pathology, concluded that 320-slice CT can provide a very useful and clear view of the cardiac structures (7). In another study, Husain S. et al. stated that the contrast-enhanced CT scan (CECT) can diagnose definitively and confirm or rule out concomitant structural abnormalities. They added that the information provided by this imaging modality has led to better therapeutic planning and an improved outcome [9]. Cardiac catheterization provides

the feasibility of single-procedure diagnosis and treatment [8]. One helpful feature of cardiac catheterization has been oximetry, which demonstrates a right-to-left shunt at the atrial level with systemic arterial desaturation. It can also reveal the elevated level of pulmonary arterial pressure (PAP), which can be seen in more severe cases, and also concomitant anomalies [15].

Surgical treatment was the preferred method of RPA-LA connection until 2000 when Slack M. et al. introduced the transcatheter treatment of this condition [16]. Similar to many other congenital or non-congenital heart anomalies, surgical replacement has been replaced with interventional strategies for this cardiovascular defect in pediatrics and adults [17, 18]. Although transcatheter treatment has been associated with risks and complications, the prominent advantages of fewer intra-operation and post-operation complications and a lower recovery time make it more favorable than the surgical option [19]. Moreover, reports of several successful transcatheter management of various anomalies make the decision-makers more interested in this option [20]. However, it is not always feasible, and sometimes surgery is necessary due to the size or structure of the malformation [14, 21]. Moreover, one of the restrictions for surgical treatment is the definite localization of the structures due to the selection of three overall thoracotomies (Median sternotomy, right posterolateral thoracotomy, or left posterolateral thoracotomy) based on the based on the accessibility of the lesion [8, 22]. Previously, there were emergencies, such as heart failure in neonates, which was the definitive indication for surgical intervention [8]. Considerable developments in transcatheter strategies have occurred, including duct occluders, septal closure devices, and vascular plugs, during recent decades, have caused the replacement of these methods for previously definite indications of surgical correction [12]. A 12×14 Amplatzer duct occluder was utilized to treat the RPA-LA fistula in a 12-year-old child through ASD after forming a venovenous loop, as by Francis et al. [23]. With a similar method, Vinothkumar S. et al. treated a 16-year-old boy (with the diagnosis of type 1 RPA-LA fistula) with a 22×24 mm Cera™ duct occluder via a transseptal approach, which resulted in an uneventful treatment and postoperative course [12].

Clinical key point (conclusion)

The diagnosis of RPA-LA is a challenge requiring high sensitivity when examining all ranges of ages. Delayed diagnosis can lead to complications such as abscess, stroke, and embolism. Considering the reduced risks associated with surgery, percutaneous intervention emerges as a suitable alternative for the treatment of this disease. However, the treatment strategies should be chosen considering the fistula's anatomical location, size, and tortuosity. It is crucial to have a precise measurement to select the most appropriate device. The usual options for percutaneous closure are duct occluder, muscular septal closure devices, and vascular plug devices.

Abbreviations

CHD	Congenital heart disease
CCHD	Cyanotic congenital heart disease
RPA-LA shunt	Right pulmonary artery to Left atrium shunt
RR	Respiratory rate
PR	Pulse rate
T	Temperature
BP	Blood pressure
CHF	Congestive heart failure
RA	Right atrium
CT	Computed tomography
CECTA	Contrast-enhanced computed tomography angiography
ECG	Electrocardiogram
CXR	Chest X-Ray
PV	Pulmonary vein
RIPV	Right inferior pulmonary vein
SOB	Shortness of breath
PFO	Patent foramen ovale
RV	Right ventricle
RA	Right atrium
PAP	Pulmonary arterial pressure
ASD	Atrial Septal Defect
TGA	Transposition of Great Arteries
TOF	Tetralogy of Fallot
VSD	Ventricular Septal Defect
LPA	Left pulmonary artery
LV	Left ventricle
EF	Ejection Fraction
TTE	Transthoracic Echocardiography
TEE	Transesophageal Echocardiography
LSVC	Left superior vena cava
LVH	Left ventricular hypertrophy
HCT	Hematocrit
LPA	Left pulmonary artery
CBP	Cardiopulmonary bypass
RPA	Right pulmonary artery
PVC	Premature ventricular contracture
RIPV	Right inferior pulmonary vein

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Author contributions
H.M., F.J., M.M., and A.T. contributed to the Conceptualization, Resource data curation and analysis, projectadministration, and writing of the initial draft. H.P. and F.N. contributed to the supervision, validation, visualization, investigation, methodology, software, and revision of the final draft of the manuscript. All authors read and approved the final manuscript.

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Data availability
Data is available on request due to privacy/ethical restrictions.

Declarations

Ethics approval and consent to participate
The study was performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. The study was approved by the Ethics Committee of Rajaei Hospital of Tehran. Considering not including any revealed information about the patient and the breach of confidentiality, the committee waived the requirement for an ethics code.

Consent statement
The patient gave written informed consent to publish this report, following the journal's patient consent policy. The procedure was performed in accordance with the center's ethical policy.

Competing interests
The authors declare no conflicts of interest.

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