

Hidden Pulmonary Arteries in Tetralogy of Fallot and Pulmonary Artery Pressure in Patients Operated with a Pulmonary Artery

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Research article

Keywords: absent pulmonary artery, tetralogy of Fallot, stent, patent ductus arteriosus

Posted Date: August 19th, 2020

DOI: <https://doi.org/10.21203/rs.3.rs-58127/v1>

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Version of Record: A version of this preprint was published on January 28th, 2021. See the published version at <https://doi.org/10.1186/s12872-021-01877-y>.

Abstract

Introduction: The absence of a pulmonary artery is a rare congenital anomaly that occurs on its own or with some congenital cardiac disorders, particularly tetralogy of Fallot (TOF), while the hidden pulmonary artery might originate from a closed ductus arteriosus (DA) that can be stented to reach the artery.

Material and methods: This prospective study describes cardiac catheterization of our nine TOF patients who had the absence of the left pulmonary artery before the operation. The patients were stratified in three groups: group one, whose closed DA were found and stented successfully to the hidden pulmonary artery; group two, whose hidden pulmonary arteries were found via the pulmonary vein angiography; and group three, for whom we could not find the remnant of the DA, or our attempt to stent the DA to the hidden pulmonary artery was not successful.

We also evaluated outcomes of the other surgically-corrected TOF patients who were operated with the absent left pulmonary artery.

Results: The first group included patients aged 1, 24, and 30 months, whose CT angiography 6-9 months after stenting showed acceptable left pulmonary artery diameter for surgical correction, while the pulmonary vein angiography of the group two patients showed a hidden left pulmonary artery with a suitable diameter for surgical correction.

However, we were unable to find or stent the DA of the group three patients, aged 12, 38, 60, and 63 months.

Moreover, evaluation of the other six previously corrected patients who were operated with a right pulmonary artery revealed pulmonary artery hypertension of the entire patients.

Conclusion: The concealed pulmonary artery might be found, and stenting of the closed DA to it might be performed to improve the diameter of the diminutive pulmonary artery. This procedure may allow TOF total surgical correction with two pulmonary arteries. Besides, pulmonary vein angiography can reveal the hidden pulmonary artery.

Introduction

Unilateral absence of a pulmonary artery (APA), also called De Buckes syndrome, is a well-known congenital heart disease with an incidence of 0.6% in patients undergoing cardiac catheterization. In total, 40% of these cases have isolated APA, and 60% suffer from other congenital cardiac disorders as tetralogy of Fallot (TOF) and pulmonary atresia with the ventricular septal defect. However, less than 3% of TOF patients have APA [1–4], and untreated APA cases might develop ipsilateral lung hypoplasia [5–7].

The trunk of the hidden pulmonary artery is occasionally absent; nonetheless, the branches of distal intrapulmonary arteries can be intact and supplied by the ductus arteriosus (DA), collateral vessels from

the subclavian, internal mammary, intercostal or other thoracic arteries [7].

Ontogenetically, the pulmonary artery is absent when the septation of a truncus is not normal. Furthermore, dorsal deviation of the right or left ridges might lead to the agenesis of the ipsilateral pulmonary artery [8, 9], and most cases develop pulmonary artery hypertension [10].

These patients have an absence of pulmonary artery origin due to the maldevelopment of the sixth aortic arch [6].

In some patients with TOF, simultaneous APA has been reported with absent pulmonary valve leaflets [11, 12].

A subset of patients has connections between DA or collaterals from bronchial, intercostal, or other arteries to the APA, hence the angiography or CT angiography of the aorta and the arteries can reveal the APA [11, 13, 14].

However, in the second subset of the cases where there is no major connection between the arteries and the hidden pulmonary artery, the APA is not visualized in imaging modalities.

In the third subset of patients, some pulmonary arteries have dual blood supplies from both the main pulmonary artery and the DA, and the pulmonary artery may have a normal or rudimentary diameter according to the stenosis of its origin (Fig. 1).

Noteworthy, after birth, the patent ductus arteriosus usually closes due to the rise in the blood oxygen level, resulting in the constriction of the smooth muscles of the DA wall; subsequently, DA degenerates to the ligamentum arteriosum, and the pulmonary artery that originates from the DA can be concealed [15–17].

In the first subset of the patients, surgical unifocalization of the pulmonary artery can be done; while, cases with undetectable pulmonary artery undergo direct connection of the right ventricle to the normal pulmonary artery [14, 18–21], leading to a lifetime with one pulmonary artery.

This study aimed to describe our experiences regarding the percutaneous finding and to open the concealed pulmonary artery and evaluate the clinical outcomes of our TOF patients with APA undergone surgical correction with one pulmonary artery branch.

Materials And Methods

This prospective study was carried out in hospitals affiliated to Shiraz University of Medical Sciences, Shiraz, Iran, 2019–2020. We introduced nine patients with a definite diagnosis of TOF and the absence of the left pulmonary artery (LPA); for whom we attempted percutaneous finding and rehabilitation of their LPA using DA stenting. Details of catheterization and stenting methods are described.

If necessary, CT angiography of the aorta and the pulmonary arteries was performed to find out the anatomy of the pulmonary arteries.

The patients were stratified in three groups according to their results of the procedures: group 1 patients, whose stumps of the DA were found, and stented toward the hidden pulmonary artery successfully; group 2 patients, whose LPAs were found via the pulmonary vein angiography which had suitable diameters for total surgical correction with no need to any intervention, and group 3 patients, whose LPAs or DA diverticulums were not found with catheterization and CT angiography.

Even though the main objective of the study was to describe finding and rehabilitation of the APA, we evaluated the outcomes of other six surgically-corrected TOF patients who were operated with the absence of the LPA, and 2-dimensional, M-mode, tissue and color Doppler echocardiography were performed to assess the left and right ventricular function and the pulmonary artery pressure.

For these patients, total correction with ventricular septal defect closure, implantation of the homograft or Contegra between the right ventricle and the main pulmonary artery, and the right pulmonary artery plication were done.

Results:

We presented nine non-operated and six operated patients from our center. The APAs were LPAs in all 15 cases. Seven patients were male and eight were female.

We decided to find and open the hidden pulmonary artery of the nine patients, which was successful in three cases. However, we could not open the pulmonary artery of the four cases, and the two other patients had a good-sized concealed pulmonary artery via pulmonary vein angiography and did not require pulmonary artery rehabilitation (Table 1).

Table 1
Demographic data of the nine non-operated patients with a hidden pulmonary

Patients (Number)	Patients (Group)	Age (Month)	Weight (kg)	Diagnosis	Aortic arch	Hidden pulmonary artery
1	Group 1	1	3.5	TOF	Right	Left
2	Group 1	24	10.5	TOF	Right	Left
3	Group 1	30	12	TOF, APV	Right	Left
4	Group 2	10	8.5	TOF	Left	Left
5	Group 2	15	9.5	TOF	Left	Left
6	Group 3	12	7.5	TOF, APV	Left	Left
7	Group 3	38	13.5	TOF	Right	Left
8	Group 3	60	15.5	TOF	Left	Left
9	Group 3	63	16	TOF	Left	Left

APV, absent pulmonary valve; F, female; M, male; Mo, month; No, number; TOF, tetralogy of Fallot

Totally eleven cases had left aortic arch, and four had the right aortic arch.

Nine DA were not in their specific place and came from underneath the aortic arch or the beginning of the brachiocephalic artery, but the DA in six cases was in its proper place.

Two patients had both APA and absent pulmonary valve.

Table 1 determines the characteristics of each non-operated nine patients, for whom we attempted to find and rehabilitate the concealed pulmonary arteries.

Group one patients, the successful group:

We described three of our patients for whom the DA finding and stenting to the absent LPA was performed successfully.

The patient one in Table 1, with arterial oxygen saturation of 55% in room air, was referred for further evaluation and possible DA stenting.

Catheterization revealed the good-sized right pulmonary artery from the right ventricle with no LPA opacification.

Aortography in anteroposterior view showed a blind-stump of the DA from the beginning of the left brachiocephalic artery towards the LPA.

A Hi-Torque Pilot coronary guidewire (Abbott Vascular) was steered from a right guiding catheter via a retrograde path through the stump of the DA to enter the LPA, and a non-compliant coronary balloon 2.5*15 millimeter was inserted and repeatedly inflated as predilation at proximal and distal parts of the stenosis. Afterward, the aortography showed the LPA opacification from the stent.

Next, a coronary stent 3*15 was inflated, and the result was promising (Fig. 2).

The patients two and three in Table 1 underwent pulmonary artery CT angiography before the catheterization which showed some small hypoplastic sparse artery branches in the left lung with no remnant of the LPA at the hilum that were non-suitable for surgical correction or palliation (Fig. 3-b).

We performed a cardiac catheterization and right ventriculography in anterior-posterior and left anterior oblique views, which showed a good-sized right pulmonary artery but no LPA opacification.

Furthermore, ascending aorta injection showed the right aortic arch and one blind-ending stump of the DA possibly towards the left lung.

We retrogradely advanced one Asahi Fielder 0.014 guidewire from a right guiding catheter through the pouch and entered the LPA. After that, we repeatedly inflated a coronary balloon with a 3*10 mm size into the stenosis; the aortic injection showed the fade staining of a small stenotic LPA from the DA.

Then, we successfully inflated one bare coronary stent size 4*18 into the DA in order to enlarge the LPA (Fig. 3-c).

Figure 3-d shows the 3-dimensional CT angiography of patient number two, six months after the procedure with an acceptable condition for TOF total correction.

Group two patients, the suitable group:

Aortography was done for group 2 patients (Table 1), which showed a closed DA stump. We decided to stent the DA but at first tried to do pulmonary vein wedge angiography to see the pulmonary artery retrogradely and to estimate the distance between the DA and the remnant of the LPA.

Luckily, the wedge angiography showed an LPA with a suitable diameter, and there was no need for rehabilitation with a stent or a surgical shunt; thus, we stopped the procedures (Fig. 4).

Group three patients, non-successful group:

The patients six and seven in Table 1 underwent CT angiography and aortography in multiple views which did not reveal the LPA or remnant of the DA (Fig. 5), while the interatrial septums were intact and, the pulmonary vein angiography was not performed due to the lack of guardian's consent for atrial septostomy.

On the other hand, the patient number 8 (and also 9) in Table 1 had a remnant of the DA, and was a 5.5-year-old girl whose cardiac catheterization at 1.5 years of age, had revealed absent LPA and a DA diverticulum; and, the total correction was carried out with right pulmonary artery and a homograft. However, she gradually developed pulmonary artery hypertension.

The previous aortic angiography was reviewed, which showed an acceptable-sized DA diverticulum, located underside of the aortic arch (Fig. 6-a). Accordingly, we decided to open the stump to the closed DA with a coronary stent.

The right axillary artery approach was chosen to insert the head of the right guiding catheter exactly in the closed DA stump. Nevertheless, aortography showed that the stump was shorter than the previous angiography.

We could not steer 0.014 guide wires Asahi Fielder, and then Pilot Abbott Vascular from the DA stump to the LPA and the end of the pouch was completely closed, and it was not possible to cross the wire (Fig. 6b - c).

Patient 9 was similar to the 8th case, hence not described for brevity.

Our operated patients

We assessed our six surgically-corrected patients other than these nine, who were operated with one pulmonary artery using a homograft or Contegra from the right ventricle to the right pulmonary artery.

The mean age of them was seven years with a range of 6 to 9 years, and mean follow-up time was four years.

All the six cases were in NYHA functional class 2–3, and echocardiography revealed severe pulmonary artery hypertension. They also developed right ventricular dysfunction while the left ventricular functions in 2-dimensional echocardiography were normal (Table 2).

Table 2
 Characteristics of the six TOF patients corrected with the absence of the left pulmonary artery

Patients (No)	Sex		F/U (year ± SD)	TR PG (mm Hg)	PS PG (mm Hg)	TAPSE (mm) and Z-score	EF%	SF%
	M	F						
6	2	4	4 ± 2.3	62	18	8 Z-score = -3.7	72	39
EF, ejection fraction; F, female; F/U, follow up; M, male; No, number; PG, pressure gradient; PS, pulmonary valvar or supra-valvar stenosis; SD, standard deviation; SF, shortening fraction; TAPSE, tricuspid annular plane systolic excursion; TOF, tetralogy of Fallot; TR, tricuspid regurgitation								

Discussion:

TOF is not only an anatomic anomaly, but it also seems to be a complicated genetic disorder that can be associated with other anomalies such as absent pulmonary valve or absence of a pulmonary artery and extracardiac anomalies [22, 23, 24].

Most patients with APA have pulmonary artery hypertension [5, 10]. A good example would be our six corrected patients who were in NYHA functional class 2 to 3, with severe pulmonary artery hypertension.

Interestingly, 65% of uncorrected patients with isolated APA who were not in TOF category developed contralateral pulmonary artery hypertension during the first three years of life [5].

Therefore, correcting the disease with two pulmonary arteries can promise a better outcome.

The right lung has more vascularity and alveolar space than the left lung [9]; therefore, patients who have the absence of the left pulmonary artery may have more favorable outcomes after surgical repair than patients with the absence of the right pulmonary artery.

Unilateral absence of the left pulmonary artery is five to eight times more frequent than the right pulmonary artery [19], which was the case with all our 15 patients who had absent left pulmonary artery while aortic arch was on the left or right sides.

Like one of our patients, some of these patients had the absence of the pulmonary valve leaflets besides APA [19].

A few studies were published regarding patients' outcomes with the absence of one pulmonary artery [5], and these cases have to spend their life with one functional pulmonary artery.

The patients 6 and 7, aged 12 and 38 months respectively, had no DA stump in the catheterization while we expected to see the DA stumps in these ages. Unfortunately, pulmonary vein angiography was not done to check the LPA, and we do not know if they had LPA or not.

Although the aortic angiography and CT scan may not detect the hidden pulmonary artery, but catheterization and particularly pulmonary vein angiography might be the preferred diagnostic method [3, 25].

The DA stenting;

A straight high tip load guidewire might be passed through the closed DA toward the concealed pulmonary artery and a stent might be inserted into the DA to rehabilitate the small PA [26]

The DA gradually changes into the ligamentum arteriosum; therefore, the sooner we perform the procedure, the more successful we might be [17, 27]. Nevertheless, we could pass the straight tip coronary guidewire through the closed DA in two 24 and 30-month-old patients.

Therefore, we do not know the exact time of the invincible closed DA.

We selected the straightest path to guide the catheter, from axillary or femoral access towards the DA, to apply maximum pressure by guidewire to the bottom of the DA pouch for penetration.

At the time of angiography, we recommend inserting an end-hole catheter into the DA diverticulum and another catheter into the pulmonary vein of the same side. The concomitant contrast agent's injection might determine the distance between the DA and the nearest part of the diminutive PA. In this way, we can select the most proper stent length.

Also, pulmonary vein wedge angiography may show pulmonary artery size and diameter. If the dimension is acceptable, we may proceed with surgical operation without intervention, as the four and five patients.

Choosing a shorter stent length may be more appropriate for reducing the risk of probing and increasing pulmonary blood flow.

Selecting a stent with a diameter equal to the related pulmonary artery diameter may prevent the hazard of the stent insertion.

Conclusion

Some patients with the impression of APA might have a concealed appropriate or diminutive-sized pulmonary artery with a closed connection between a DA and the pulmonary artery. In this regard, a precise aortic arch angiography might show a DA stump that predicts a connection between DA and the adjacent pulmonary artery. Early stenting may be more successful, and PA rehabilitation can improve pulmonary blood circulation and lung development.

Limitation Of The Study

Pulmonary vein angiographies could not be done in two our patients whose DA stumps were not seen in the aortic arch injections, and we suggest doing this procedure to distinguish between an actual APA and a concealed pulmonary artery to prevent any misunderstanding.

Abbreviations

APA: Absence of a pulmonary artery

DA: Ductus arteriosus

LPA: Left pulmonary artery

TOF: Tetralogy of Fallot

Declarations

Ethics approval and consent to participate:

The Ethics Committee of Shiraz University of Medical Sciences has approved this study (the ethical code: IR.SUMS.med.rec.1398.568). All procedures comply with the ethical standards of the relevant national guidelines on human experimentation of institutional and ethics committees. Furthermore, the study was according to the Helsinki Declaration of 1975 and its revised version in 2008. For all parents or guardians, the study was explained, and written consent was taken.

Consent for publication:

this manuscript does not contain any personal data, and the consent for publication is applicable, and the patient guardians gave us the consent for these clinical and image data to be published in this study.

Availability of data and material:

also, concerning data availability, we state that the data used and analyzed during the current study are available from the corresponding author on reasonable request. Data sharing applies to this article, and datasets were generated and analyzed during the current study, and data sharing is allowed.

Conflict of interest:

none declared, and the authors did not receive any direct or indirect financial payment for the research and are not owners of any related company and are not consultants of these companies.

Funding:

there was no any funder for this research.

Author contributions:

ME: Design and writing the manuscript

K.K.: Corresponding author

A.A.: Operation

B.G.: Operation

H.A.: Echocardiography

Z.K.: Echocardiography

H.M.: Data collection

GA: Critical revision

NM: Data collection

AN: Manuscript revision

F.P.: Manuscript revision

SC: Echocardiography

MN: Data collection

All authors have read and approved the manuscript.

Acknowledgment:

The authors would like to thank Shiraz University of Medical Sciences, Shiraz, Iran, for financial support. The authors wish to thank Mr. H. Argasi at the Research Consultation Center (R.C.C.) of Shiraz University of Medical Sciences for his invaluable assistance in editing this manuscript.

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Figures

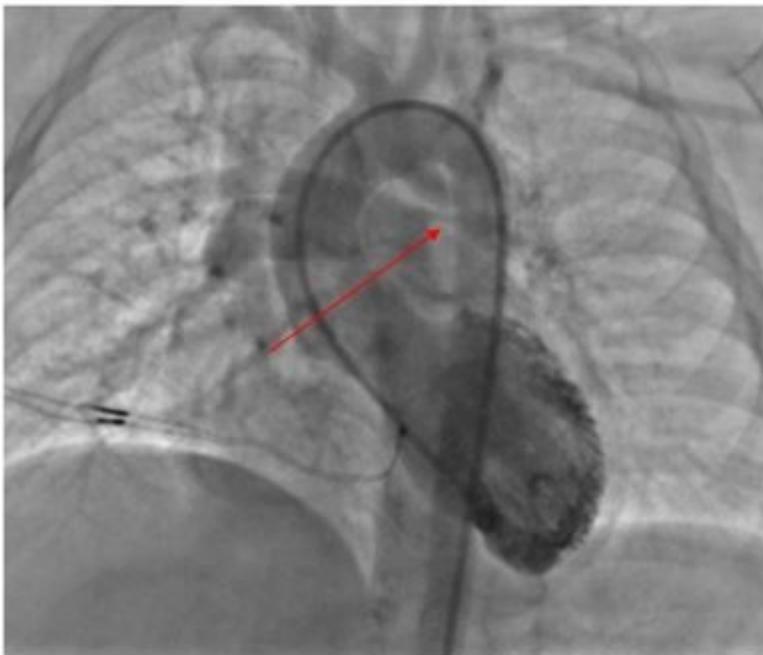


Figure 1

Double supply diminutive left pulmonary artery. The arrow shows the origin of the stenotic pulmonary artery.

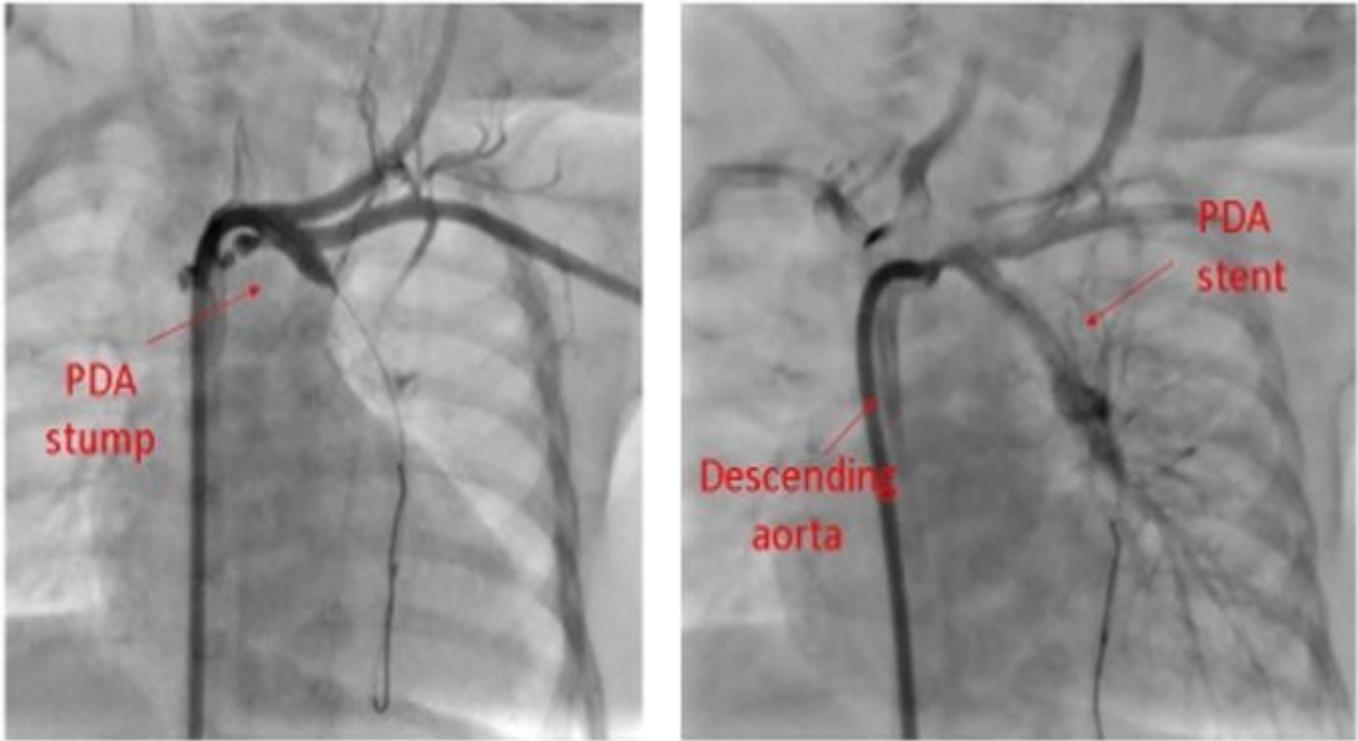


Figure 2

Stenting of the ductus arteriosus towards the left pulmonary artery

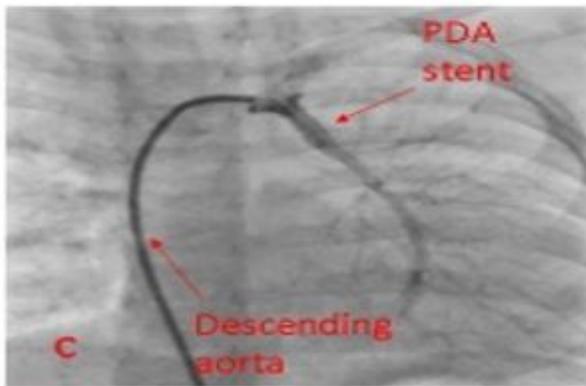
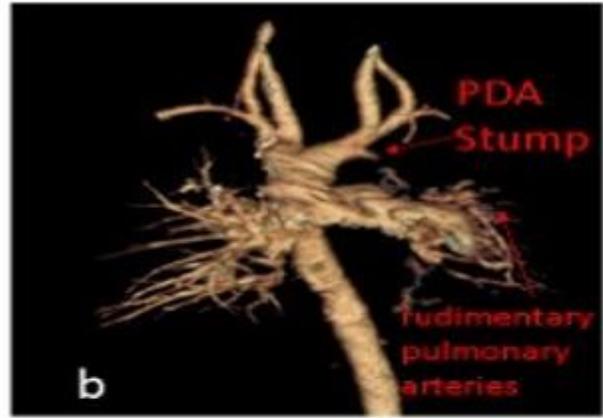


Figure 3

Successful stenting of the ductus arteriosus to the left pulmonary artery in our third patient; a, right ventriculography; b, C.T. angiography before the procedure; c, stenting of the left pulmonary artery; d, C.T. angiography six months after the procedure

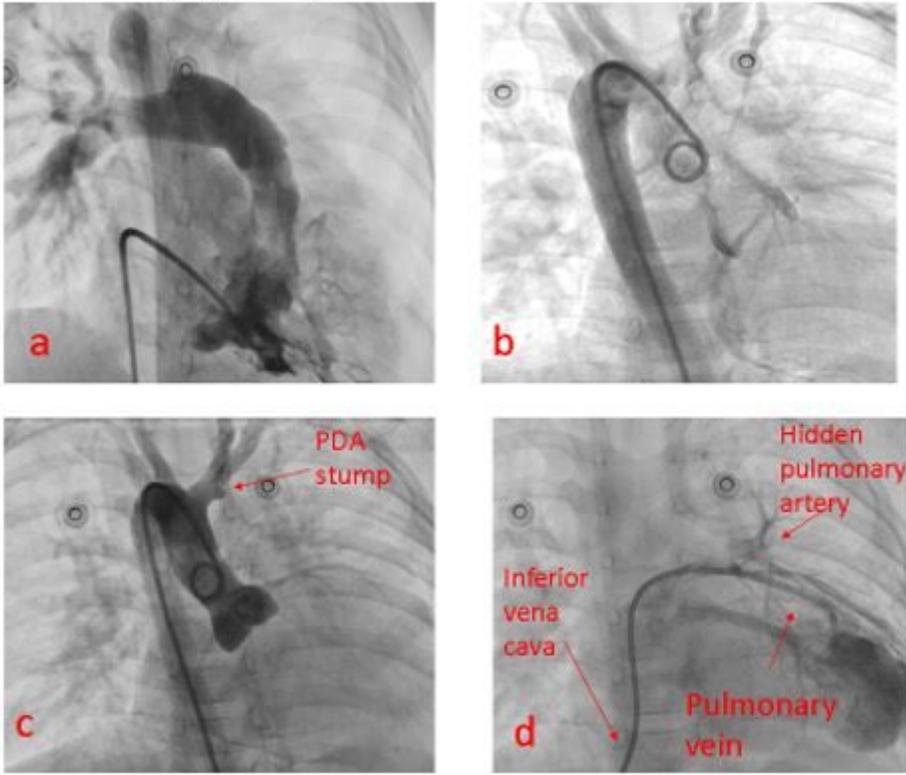


Figure 4

absent LPA. B: aortography shows no collateral. C: the arrow shows a ductus arteriosus pouch. D: the hidden left pulmonary artery following pulmonary vein wedge angiography.

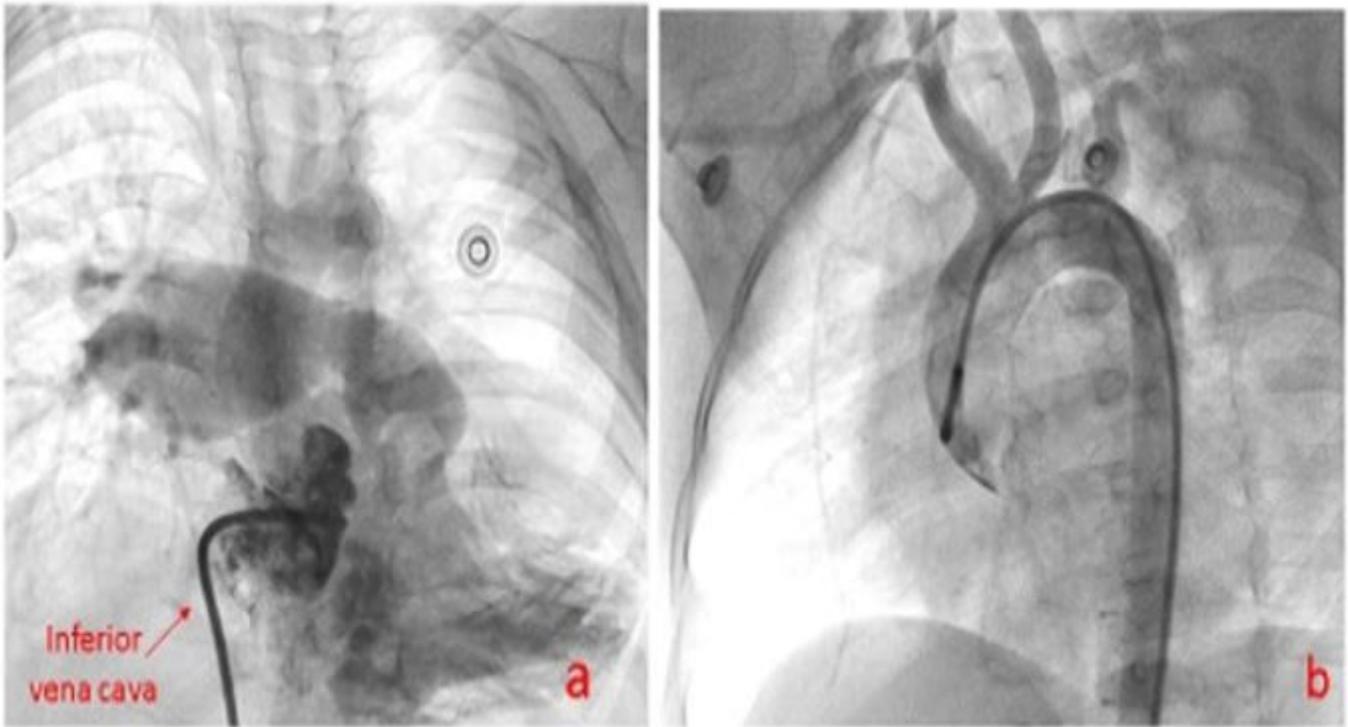


Figure 5

Absence of the left pulmonary artery in patient with no remnant of the ductus arteriosus; a, right ventricle catheterization; b, aortography.

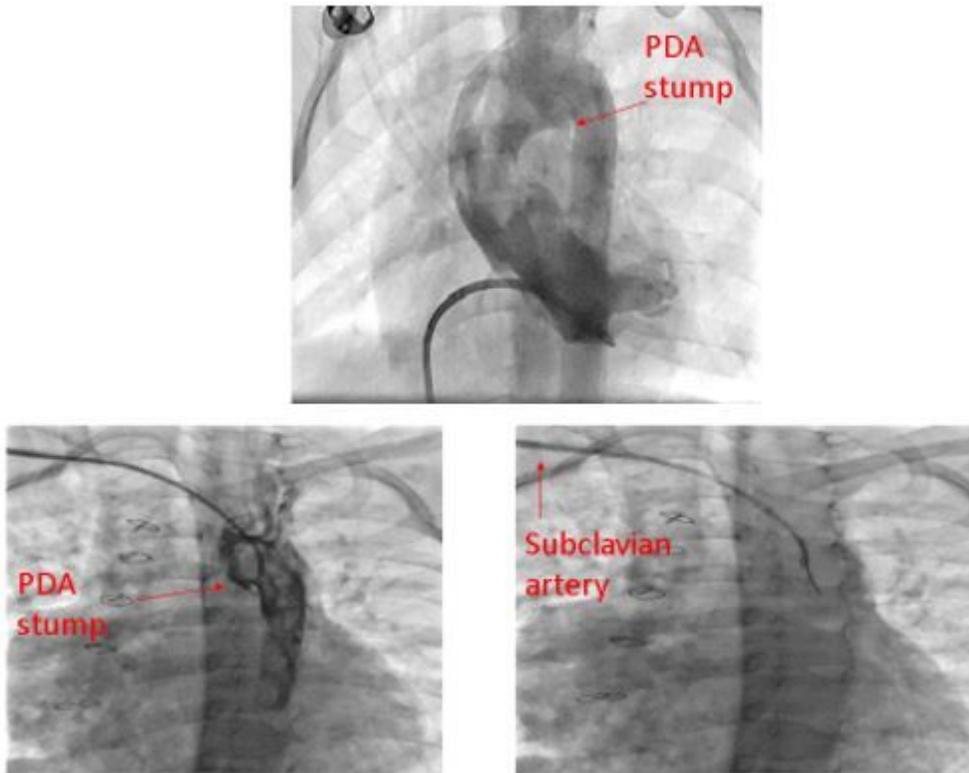


Figure 6

absent LPA. B: aortography shows no collateral. C: the arrow shows a ductus arteriosus pouch. D: the hidden left pulmonary artery following pulmonary vein wedge angiography.