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Anomalous connection of the circumflex coronary artery to the pulmonary trunk in a patient with Taussig–Bing anomaly: a case report

Jef Van den Eynde ^{1,2}, Lukas A.J. Rammeloo³, Monique R.M. Jongbloed ^{4,5}, Mark G. Hazekamp ¹, and Roel L.F. van der Palen ^{6*}

¹Department of Cardiothoracic Surgery, Leiden University Medical Center, Albinusdreef 2, 2333 ZA, Leiden, The Netherlands; ²Department of Cardiovascular Diseases, KU Leuven, Herestraat 49, 3000 Leuven, Belgium; ³Department of Pediatrics, Division of Pediatric Cardiology, Amsterdam UMC, Vrije Universiteit Amsterdam, Meibergdreef 9, 1105 AZ, Amsterdam, The Netherlands; ⁴Department of Cardiology, Leiden University Medical Center, Albinusdreef 2, 2333 ZA, Leiden, The Netherlands; ⁵Department of Anatomy & Embryology, Leiden University Medical Center, Albinusdreef 2, 2333 ZA, Leiden, The Netherlands; and ⁶Department of Pediatrics, Division of Pediatric Cardiology, Leiden University Medical Center, Albinusdreef 2, 2333 ZA, Leiden, The Netherlands

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Background

Coronary anomalies are present in one-third of all patients with transposition of the great arteries (TGA) and have been associated with increased risk of adverse outcomes after the arterial switch operation. Therefore, knowledge about coronary anatomy remains key.

Case summary

A 5-day-old girl with prenatal diagnosis of Taussig–Bing anomaly (double outlet right ventricle with TGA and large subpulmonary ventricular septal defect) along with aortic arch hypoplasia and coarctation of the aorta underwent the arterial switch operation with closure of the ventricular septal defect and aortic arch repair. On preoperative echocardiography, the right (R) and left coronary artery (LCx) connected both to aortic sinus 1, suggesting 1RLCx coronary anatomy according to the Leiden Convention coronary coding system. However, intraoperative inspection led to a reclassification of the coronary anatomy: the right coronary artery and left anterior descending coronary artery connected to aortic sinus 1 (1RL) as had been observed on echocardiography, but—remarkably—the circumflex coronary artery (Cx) connected to the posterior sinus of the pulmonary trunk. As a consequence, cardioplegia was administered into both the aortic and pulmonary roots, and the circumflex coronary artery could stay in its native position without having to be transferred during the arterial switch operation.

Discussion

Various disruptions during embryological development can lead to unusual coronary anatomy in TGA patients. While anomalous connection of a coronary artery to the pulmonary trunk remains exceedingly rare, care should be taken to identify this pattern when present as failure to do so may result in adverse outcomes.

Keywords

Arterial switch operation • Case report • Congenital heart disease • Coronary artery • Echocardiography • Transposition of the great arteries

ESC Curriculum

9.7 Adult congenital heart disease • 3.1 Coronary artery disease • 2.2 Echocardiography • 2.1 Imaging modalities • 7.5 Cardiac surgery

* Corresponding author. Tel: +31 715 262 835, Fax: +31 715 248 110, Email: r.vanderpalen@lumc.nl

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Learning points

- While very rare in nature, failure to recognize anomalous connection of a coronary artery to the pulmonary trunk in patients with transposition of the great arteries can have potentially devastating consequences.
- Thorough preoperative planning, meticulous intraoperative inspection, and careful postoperative monitoring with regard to coronary anatomy are keys in all patients undergoing the arterial switch operation.

Introduction

Outcomes after the arterial switch operation for dextro-transposition of the great arteries (TGA) have improved considerably, yet knowledge about the coronary anatomy remains essential.¹ Coronary anomalies are present in about one-third of all patients with TGA, and even more frequently in certain subgroups such as Taussig–Bing anomaly.² Unusual coronary patterns have been associated with increased risk of adverse postoperative outcomes, and some of these patterns have specific implications for management.^{2,3} Therefore, thorough preoperative planning, meticulous inspection in the intraoperative setting, and careful postoperative monitoring with regard to coronary anatomy are key in all patients undergoing the arterial switch operation. Here, we describe a case of a newborn girl with Taussig–Bing anomaly in whom an anomalous connection of the circumflex coronary artery (Cx) to the pulmonary trunk was identified.

Timeline

Age	Event
27 weeks' gestational age	Prenatal diagnosis of critical congenital heart disease: Taussig–Bing anomaly [double outlet right ventricle (DORV) with transposition of the great arteries and a large subpulmonary ventricular septal defect (VSD)], aortic arch hypoplasia, coarctation of the aorta.
0 days	Birth at 39 weeks and 5 days' gestational age (birth weight 3.0 kg), with subsequent transfer to the Neonatal Intensive Care Unit for preoperative care. Postnatal echocardiography suggests 1RLCx coronary anatomy according to the Leiden Convention coronary coding system.
5 days	Arterial switch operation with VSD patch closure and aortic arch repair. Intraoperative inspection led to a reclassification of the coronary anatomy: the circumflex coronary artery connected to the posterior sinus of the pulmonary trunk. Therefore, only the right coronary artery and left anterior descending coronary artery needed transferal to the neo-aorta (native pulmonary trunk).
22 days	Discharged home.
1–10 months	Development of progressive valvular and supravalvular right ventricular (RV) outflow tract obstruction (RVOTO).

Continued

Continued

Age	Event
10 months	Failed balloon dilatation of the RVOTO.
11 months	Successful surgical relief of the RVOTO using a transannular patch. Preoperative computerized tomography angiography showed patency of the reimplanted (RL) coronary artery button as well as the native circumflex coronary artery.
28 months	Patient is doing well with good biventricular systolic function.

Case presentation

A female newborn, delivered at 39 weeks and 5 days' gestational age (birth weight 3.0 kg) was delivered by means of planned caesarean section at our tertiary referral centre, given the antenatal diagnosis of a critical congenital heart disease. Foetal echocardiography had revealed DORV with TGA and a large subpulmonary VSD, indicating Taussig–Bing anomaly, along with aortic arch hypoplasia and coarctation of the aorta. Postdelivery, the neonate was admitted to the neonatal intensive care unit without respiratory and circulatory support and stable saturations of 85%. To maintain ductal patency in the setting of aortic coarctation, prostaglandin E2 was initiated.

Postnatal echocardiography revealed a size discrepancy between the great arteries with a larger pulmonary artery compared with the aorta: aortic annulus (8.2 mm, Z-score 1.9) in a right anterior position compared with the enlarged pulmonary annulus (12.3 mm, Z-score 3.1).⁴ While the ascending aorta was normal sized (7.5 mm, Z-score –0.03), there was hypoplasia of the proximal aortic arch (5.9 mm, Z-score –1.8), isthmus (2.5 mm, Z-score –4.7), and distal aortic arch (3.2 mm, Z-score –3.8).⁴ Apart from the prenatally diagnosed subpulmonary perimembranous VSD, two additional small apical VSDs were identified and there was a good communication at atrial septal level with left-to-right shunt, not necessitating balloon atrial septostomy. The right and left coronary arteries seemed to connect both to aortic sinus 1 from the same ostium, suggesting 1RLCx coronary anatomy according to the Leiden Convention coronary coding system (Figure 1A).⁵ The anatomical diagnosis was deemed sufficiently conclusive from echocardiography, such that a computerized tomography angiography (CTA) was not performed at this point.

On postnatal Day 5, the patient underwent arterial switch operation including Lecompte manoeuvre, VSD patch closure, and aortic arch repair. Intraoperative inspection led to a reclassification of the coronary anatomy: the right coronary artery (R) and left anterior descending coronary artery (L) connected to aortic sinus 1 (1RL) as had been observed on echocardiography, but—in contrast to the initial diagnosis—the Cx connected to the posterior sinus of the pulmonary trunk (Figure 2). As a consequence, cardioplegia was administered into both the aortic and

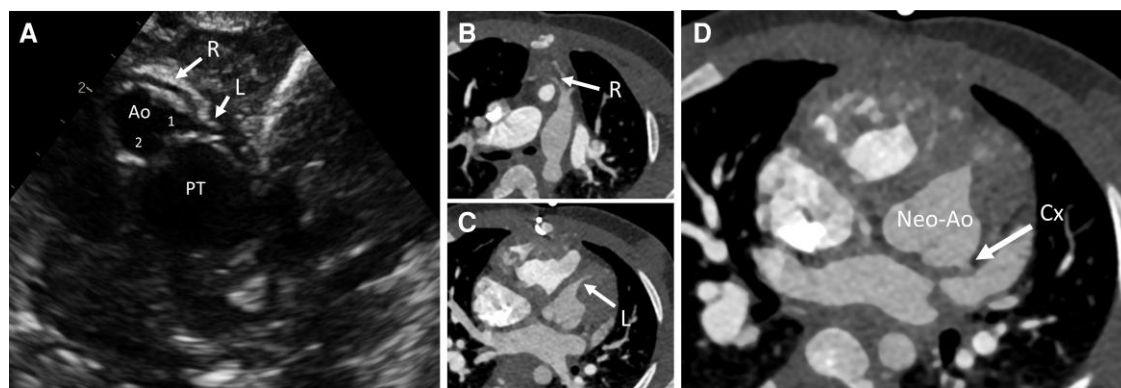


Figure 1 Coronary artery distribution pattern in our patient. (A) Postnatal transthoracic echocardiography showing the connection of the R and L to aortic sinus 1 from the same ostium, suggesting 1RLCx coronary anatomy according to the Leiden Convention coronary coding system. (B–D) Preoperative computerized tomography angiography prior to right ventricular outflow tract relief showing patency of the reimplanted (RL) coronary artery button in the anterior position of the neo-aortic sinus (B and C) and the circumflex coronary artery at its native connection to the posterior position of the neo-aortic sinus (D). The latter had only been noticed during the arterial switch operation, resulting in reclassification to 1RL coronary anatomy with anomalous connection of the circumflex coronary artery to the pulmonary trunk. Ao, native aorta; CTA, computerized tomography angiography; Cx, circumflex coronary artery; L, left anterior descending coronary artery; Neo-Ao, neo-aorta; PT, pulmonary trunk; R, right coronary artery; RVOT, right ventricular outflow tract.

pulmonary roots, and the Cx could stay in its native position without having to be transferred to the neo-aorta (i.e. the native pulmonary trunk). The operation was uncomplicated; the postoperative course was notable for two episodes of pulmonary hypertensive crisis from

which the patient recovered well following treatment with supplemental oxygen, fluid resuscitation, adrenaline (at maximum dose of 0.09 µg/kg/min intravenously; in addition to milrinone 0.50 µg/kg/min and noradrenaline 0.22 µg/kg/min intravenously), deep sedation, and neuromuscular

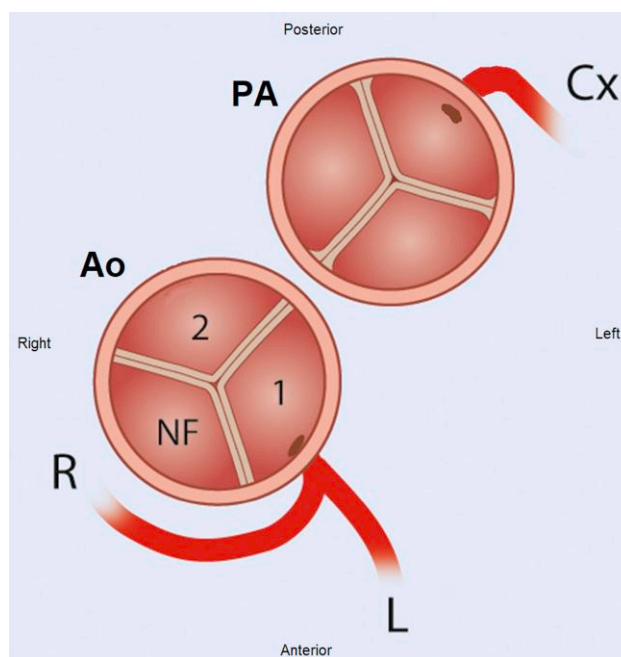


Figure 2 Schematic representation of the actual native coronary artery distribution pattern in our patient, view from above. The R and L connect to aortic sinus 1, while the circumflex coronary artery connects to the posterior sinus of the pulmonary valve. Modified based on Figure 4D from Gittenberger-de Groot et al.⁵ Ao, native aorta; Cx, circumflex coronary artery; L, left anterior descending coronary artery; PT, pulmonary trunk; NF, non-facing sinus; R, right coronary artery.

Table 1 Classification of coronary artery anomalies

Type of anomaly	Variant	Comment
Anomalies of connection	Anomalous connection of the coronary artery to the pulmonary trunk	Subtypes include those involving the left main, right, circumflex, or combined left and right coronary artery
	Anomalous connection of the coronary artery to the aorta	Subtypes include various combinations involving anomalous connections to the right or left aortic sinus of Valsalva, connections of coronary arteries to one another, single coronary arteries, and inverted coronary arteries
	Congenital atresia of the left main artery	Rare cause of myocardial ischemia and sudden death
Anomalies of course	Retroaortic/interarterial/subpulmonic/prepulmonic/retrocardiac course	Some of these are to be considered 'malignant' due to increased risk of myocardial ischemia and sudden death from dynamic obstruction/occlusion
	Myocardial (or coronary) bridging	Identified on 0.5–12% invasive coronary angiographies
	Coronary aneurysm	Usually acquired due to various systemic diseases (e.g. atherosclerosis, syphilis, Kawasaki disease), while congenital cases are extremely rare
Anomalies of termination	Coronary arteriovenous fistula	Identified in up to 0.9% of children with complex congenital heart disease, but rare in adults
	Coronary stenosis	

Adapted and modified from Gentile et al.¹⁰

blocking agents (midazolam 0.1 mg/kg/h, morphine 15 µg/kg/h, and rocuronium 1.6 mg/kg/h intravenously; doses are those maximally administered). After 7 days at the paediatric intensive care unit, the patient could be transferred to the paediatric cardiology ward, and 10 days later, she could be discharged home.

During follow up, the patient developed progressive valvular and supraventricular right ventricular outflow tract obstruction (RVOTO), increasing RV pressures that reached systemic pressures, and RV hypertrophy. Following failed balloon dilatation of the obstruction (no change in gradient obtained) at the age of 10 months, the patient underwent successful surgical RVOTO relief using a constrained transannular patch at the age of 11 months, carefully taking into account the

right coronary artery crossing the RVOT. Both procedures were uncomplicated. The CTA obtained prior to the reoperation is presented in [Figure 1B–D](#), showing patency of the RL coronary artery button in the anterior position of the neo-aortic sinus ([Figure 1B and C](#)) and the Cx at its native posterior position in the neo-aortic sinus ([Figure 1D](#)).

At latest follow up at the age of 28 months, our patient is doing well with good biventricular systolic function. The residual RVOT gradient and moderate neo-pulmonary regurgitation, as a result of the transannular patch for RVOTO relief, are closely monitored but have not required reintervention thus far.

Discussion

We report a case of anomalous connection of the Cx to the pulmonary trunk in a patient with Taussig–Bing anomaly. In normally related great vessels, the occurrence of anomalous connection of a coronary artery to the pulmonary trunk (ACAPT) is only 1 in 10 550 and usually involves the left coronary artery (in 83% of the cases).⁶ Although TGA is associated with a variety of coronary anomalies in about one-third of the patients,^{3,5} ACAPT has been reported in only three cases before, including two cases involving the left coronary artery^{7,8} and one involving the right coronary artery.⁹ The latter was, as in our case, a patient with Taussig–Bing anomaly. Our present case report is therefore the second to demonstrate ACAPT in a patient with Taussig–Bing anomaly and the first to report ACAPT involving the Cx in patients with TGA. Apart from ACAPT, a number of other coronary artery anomalies have been identified and are classified according to connection, course, and termination of the coronary arteries.¹⁰ A summary is provided in [Table 1](#), but a more detailed discussion of the epidemiology, diagnosis, treatment, and prognosis of each of these variants is beyond the scope of this article and has been provided elsewhere.¹¹

Various explanations have been proposed regarding the development of ACAPT. One of the earliest theories, by Abrikosoff in 1911,¹² suggests a role for abnormal septation of the outflow tract into the aortic and pulmonary trunks. The left aortic sinus lies close to the aortopulmonary septum, and it can easily be imagined how a small relative displacement of the site of septation could lead to the inclusion of the left coronary ostium into the pulmonary sinus. The observation that aortopulmonary window is commonly associated with ACAPT involving the left coronary artery seems to support abnormal septation as a common embryological ground for both anomalies.¹³ However, the theory by Abrikosoff falls short in the face of other variants, such as those involving the right coronary artery or Cx.

Alternatively, the Hackensellner involution-persistence hypothesis proposes that all six semilunar valve regions of the aorta and pulmonary trunk have the propensity to develop coronary buds.¹⁴ The various forms of ACAPT can then be explained on the basis of faulty involution or persistence of one or several of these coronary buds. Research from the late 1980s by the Leiden group of Prof. Dr Gittenberger-de Groot led to the conception that the coronary arteries do not grow out of the aorta, but grow into the aorta from the peritruncal ring of coronary arterial vasculature.¹⁵ This provided explanations for the coronary artery variations in TGA and common arterial trunk. Later research in chick models showed that adequate coronary ingrowth likely requires signalling from epicardial cells.¹⁶ The current understanding is that persistence or involution of the coronary buds is driven by the presence or absence, respectively, of signals from the coronary arteries and their surroundings.¹¹ This also allows to better understand the embryological origins of ACAPT, whereby abnormal ingrowth in the arterial pole leads to abnormal connections of coronary arteries to the pulmonary trunk. In this regard, it is more appropriate to use the newer terminology of 'coronary artery connected to the pulmonary trunk' rather than its historical version 'origin of the coronary artery from the pulmonary artery'.

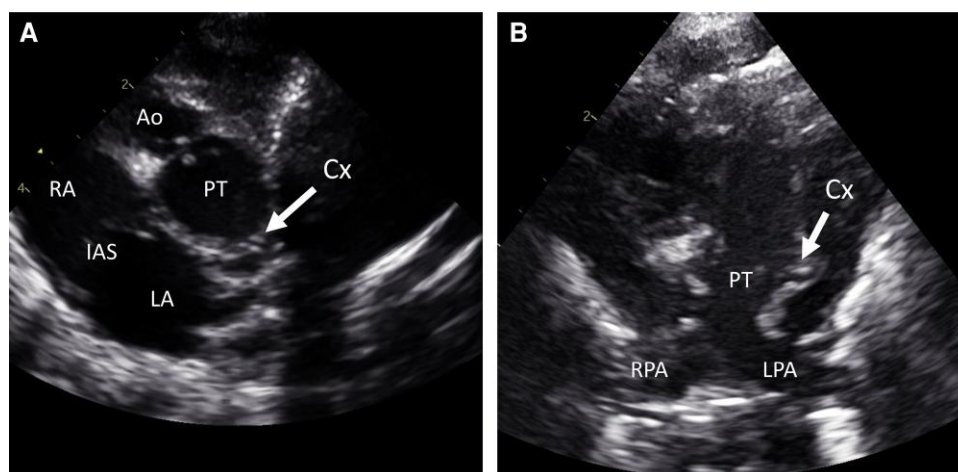


Figure 3 Circumflex coronary artery on postnatal transthoracic echocardiography. Upon retrospective review of the postnatal transthoracic echocardiography images led to the identification of a structure that could have been recognized as the circumflex coronary artery connecting to pulmonary trunk at the time of acquisition, but was likely missed due to the rarity of this feature. Transthoracic echocardiographic short axis view of the great arteries (A) and a rotated transthoracic echocardiographic view of the pulmonary trunk (B). Ao, aorta; Cx, circumflex coronary artery; IAS, interatrial septum; LA, left atrium; LPA, left pulmonary artery; PT, pulmonary trunk; RA, right atrium; RPA, right pulmonary artery.

The coronary anomaly in our patient was initially missed on postnatal echocardiography. Upon retrospective review of these images, a structure was identified that could have been recognized as the Cx connecting to the pulmonary trunk at the time of acquisition, but was likely missed due to the rarity of this condition and because the Cx in some neonates with TGA cannot be visualized anyways (Figure 3A and B). It could be suggested that a higher probability of identifying the anomalous Cx might have been obtained if preoperative CTA was also performed. However, CTA in neonates presents a major practical challenge due to their faster heart rates, small vessel size, and inability to lie still or breath-hold (with resulting need of sedation). Furthermore, echocardiography can accurately diagnose coronary anatomy in the majority of cases, including the more common cases of ACAPT involving the left or right coronary artery.¹⁷ Thus, echocardiography remains the gold standard imaging modality for coronary assessment prior to the arterial switch operation, while CTA is reserved for selected TGA cases prior to the arterial switch operation (i.e. those with suspected complex coronary anatomy such as interarterial and intramural coronary arteries, those with large coronary fistulae, and those with unclear coronary anatomy due to poor acoustic windows) and for screening for coronary complications after the arterial switch operation. As our present case suggests, suspicion of unusual types of ACAPT (e.g. non-identifiable Cx) might also serve as an indication for CTA, taking into account the limitations and practical challenges related to CTA in neonates and considering that adequate intraoperative inspection usually serves as a ‘safety net’ for these cases even if CTA is not performed.

Fortunately, the anomalous connection of the Cx was promptly identified intraoperatively, as this has some major implications for the arterial switch operation. First, myocardial protection in any type of cardiac surgery is a key, and failure to identify ACAPT will result in ischaemia with potentially devastating results. Therefore, cardioplegia had to be administered directly into both the aortic and pulmonary roots in our patient. Second, the arterial switch procedure is actually simplified in the presence of ACAPT, since the anomalous coronary artery does not have to be transferred. Finally, care should be taken to identify the location of the anomalous orifice of the ACAPT prior to

transecting the pulmonary trunk, because it may have a high take-off distally relative to the pulmonary sinotubular junction.¹³

Conclusions

In conclusion, this case report demonstrated anomalous connection of the Cx to the pulmonary trunk in a patient with Taussig–Bing anomaly. In the setting of TGA, ACAPT presents some particular challenges and implications for diagnosis and management around the arterial switch operation. Careful and thorough preoperative assessment of unusual coronary anatomy is a key to ensure optimal outcomes in this unique patient population.

Lead author biography



Jef Van den Eynde is a final year medical student and researcher at the KU Leuven (Belgium), Research Fellow at the Johns Hopkins Hospital (USA), and during the conduct of the present work was completing a clinical internship at the Leiden University Medical Center (The Netherlands). At the age of 23, Jef has already published >100 peer-reviewed manuscripts, served as a reviewer, and editor for several journals, and been a speaker at conferences all over the world. Although he keeps a broad interest in cardiovascular medicine, he has committed himself to improving the outcomes of those with congenital heart disease.

Supplementary material

Supplementary material is available at *European Heart Journal – Case Reports* online.

Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as [Supplementary data](#).

Consent: The authors confirm that written consent for submission and publication of this case report including images and associated text has been obtained from the parents of this patient in line with COPE guidance.

Conflict of interest: None declared.

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