

# Polish Heart Journal

The Official Peer-reviewed Journal of the Polish Cardiac Society since 1957

# **Online first**

This is a provisional PDF only. Copyedited and fully formatted version will be made available soon

ISSN 0022-9032 e-ISSN 1897-4279

### Congenital coronary aneurysm and cameral fistula embolization in a teenager

Authors: Jacek Kuźma, Mariusz Kuśmierczyk, Krzysztof Godlewski, Remigiusz Krysiak,

Jarosław Żyłkowski, Michał Buczyński

Article type: Clinical vignette

Received: July 30, 2023

Accepted: August 27, 2023

Early publication date: September 1, 2023

This article is available in open access under Creative Common Attribution-Non-Commercial-No Derivatives 4.0 International (CC BY-NC-ND 4.0) license, allowing to download articles and share them with others as long as they credit the authors and the publisher, but without permission to change them in any way or use them commercially.

Congenital coronary aneurysm and cameral fistula embolization in a teenager

**Short title:** Coronary aneurysm embolization

Jacek Kuźma<sup>1</sup>, Mariusz Kuśmierczyk<sup>1</sup>, Krzysztof Godlewski<sup>2</sup>, Remigiusz Krysiak<sup>3</sup>, Jarosław

Żyłkowski<sup>4</sup>, Michał Buczyński<sup>1</sup>

<sup>1</sup>Department of Cardiothoracic and Transplantology, Medical University of Warsaw,

Warszawa, Poland

<sup>2</sup>Pediatric Cardiology and General Pediatrics Clinic, Medical University of Warsaw, Warszawa,

Poland

<sup>3</sup>Department of Pediatric Radiology, Medical University of Warsaw, Warszawa, Poland

<sup>4</sup>2<sup>nd</sup> Department of Clinical Radiology Medical University of Warsaw, Warszawa, Poland

**Correspondence to:** 

Jacek Kuźma, MD.

Department of Cardiothoracic and Transplantology,

Medical University of Warsaw,

Żwirki i Wigury 63A, 02–091 Warszawa, Poland,

phone: +48 22 317 98 81,

e-mail: jacek.kuzma@wum.edu.pl

Coronary interventions in children are rare and mostly due to congenital or acquired anomalies

including abnormal origin from pulmonary trunk, coronary and cameral fistulas, vasculitis

(e.g., Kawasaki and Takayasu diseases or systemic lupus erythematosus with aneurysm

development) [1–3] or cardiac surgeries with coronary transplantation.

A 17-year-old girl was reffered for cardiac evaluation due to heart murmur found on

auscultation in a pediatric office. In medical history, the patient was asymptomatic without

symptoms of chest pain or easy fatigability. A diagnosis of right coronary artery (RCA)

aneurysm with a cameral fistula was established in a cardiology department in transthoracic

echocardiography (TTE), computed tomography and coronary angiography with negative

treadmill exercise stress test. The patient was referred for heart surgery. On admission to the

cardiac surgery center the vital signs were normal with regular heart rate of 72 beats per minute

and blood pressure of 100/60 mm Hg. Myocardial biomarkers (NT-proBNP and troponin) were

within normal range. Electrocardiogram (ECG) showed normal sinus rhythm without features of ventricular hypertrophy or myocardial ischemia. TTE revealed normal myocardial contractility, dilated proximal RCA (6 mm) with turbulent flow over the right ventricular (RV) wall. The child was qualified for initial interventional RCA aneurysm embolization and in case of failure, surgery was an option.

Aortic root angiography showed proximal RCA dilation, critical pre-aneurysmal stenosis (1mm), large right coronary aneurysm ( $10 \times 7$  mm) with cameral fistula stealing the blood into the RV (Figure 1A–C, Supplementary materials, *Videos S1–S3*). The aneurysm continued into the distal RCA supplied abundantly from circumflex artery (Cx) collateral circulation. A balloon occlusion test of RCA aneurysm with a 4mm Tyshak balloon catheter was performed with simultaneous ECG evaluation. It showed normal ECG tracings indicating sufficient Cx collateral circulation.

Arterio-venous wire loop was established with multipurpose catheter over 0.014-inch guidewire and an Amplatz 6 mm Goose Neck<sup>TM</sup> snare system (ev3, Plymouth, MN, US) by crossing the aorta, right coronary aneurysm, cameral fistula, RV and inferior vena cava (Supplementary materials, *Videos S4–S7*). Unfortunately, the access to the aneurysm with a 4 F multipurpose catheter via cameral fistula was inapplicable due to small size of the fistula (less than 2 mm).

Finally, the approach through the aortic root and critically RCA stenosis was established with a 2.9 F catheter. The aneurysm was successfully embolized with neurological detachable Penumbra Coil 400 system and PAC coils (Penumbra, Alameda, CA, US) (Figure 1D–F, Supplementary materials, *Videos S8–S11*). The clinical course was uneventful with normal ECG and myocardial contractility in TTE. The troponin level was transiently elevated up to 90 ng/l (normal range < 26.2 ng/l) with normalization within 3 days.

In 1 year follow up the girl was in a good condition with normal TTE and MRI (LVEF 60%, RVEF 59%) without features of myocardial ischemia.

In conclusion the authors underline that percutaneous or hybrid coronary interventions became alternative options in children with coronary abnormalities [4]. The dilemma of embolizing RCA in case of life-threatening aneurysm may be resolved with a balloon occlusion test and LCA angiography showing sufficient collateral circulation [5].

### **Supplementary material**

Supplementary material is available at https://journals.viamedica.pl/kardiologia\_polska.

#### **Article information**

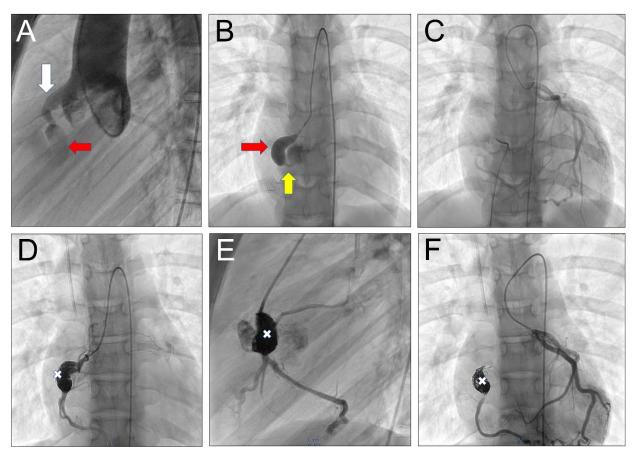
Conflict of interest: None declared.

Funding: None.

**Open access:** This article is available in open access under Creative Common Attribution-Non-Commercial-No Derivatives 4.0 International (CC BY-NC-ND 4.0) license, which allows downloading and sharing articles with others as long as they credit the authors and the publisher, but without permission to change them in any way or use them commercially. For commercial use, please contact the journal office at kardiologiapolska@ptkardio.pl.

### **REFERENCES**

- 1. Feng J, Zhao J, Li J, et al. Classification, diagnosis and clinical strategy of congenital coronary artery disease in children. Front Pediatr. 2023; 11: 1132522, doi: 10.3389/fped.2023.1132522, indexed in Pubmed: 36969282.
- 2. Lee M, Meidan E, Son M, et al. Coronary artery aneurysms in children is not always Kawasaki disease: a case report on Takayasu arteritis. BMC Rheumatol. 2021; 5(1): 27, doi: 10.1186/s41927-021-00197-0, indexed in Pubmed: 34380576.
- 3. Posadzy-Małaczyńska A, Woźnicka-Leśkiewicz L, Juszkat R, et al. Right coronary artery aneurysm with fistula into the coronary sinus in patient with systemic lupus erythematosus. Kardiol Pol. 2013; 71(12): 1329, doi: <a href="https://doi.org/10.5603/KP.2013.0341">10.5603/KP.2013.0341</a>, indexed in Pubmed: <a href="https://doi.org/10.5603/KP.2013.0341">24399601</a>.
- 4. Pająk J, Karolczak MA, Buczyński M, et al. Coronary steal phenomenon following right ventricle decompression and revascularization of atretic left coronary ostium: case report. J Cardiothorac Surg. 2021; 16(1): 299, doi: 10.1186/s13019-021-01681-x, indexed in Pubmed: 34645497.
- Kuźma J, Weryński P, Skorek P, et al. Critical value of the balloon occlusion test of a coronary fistula in a patient with pulmonary atresia and intact ventricular septum (RCD code: I 1C.4; II 2A.1). J Rare Cardiovasc Dis. 2020; 4(3), doi: 10.20418/jrcd.vol4no3.357.



**Figure 1. A**. Aortography in lateral view showing right coronary proximal dilation (white arrow), pre-aneurysmal stenosis and large coronary aneurysm (red arrow). **B.** Selective right coronary angiography (antero-posterior view) showing large aneurysm (red arrow) and cameral fistula (yellow arrow) draining the blood into the right ventricle. **C.** Selective left coronary angiography (antero-posterior view) showing collateral circulation with right coronary artery. **D** and **E.** Selective right coronary aneurysm embolized with Penumbra coils (white cross). **F.** Selective left coronary angiography (antero-posterior view) showing collateral circulation without steal phenomenon via embolized cameral fistula