

Comment on Aortopulmonary Window and Anomalies of Coronary Arterial Origin


Sir,

In 2 recent case reports published in the journal, attention has been drawn to the association between an aortopulmonary window and anomalous origin of the left coronary artery from the pulmonary trunk.^{1,2} In the manuscript recently listed as an “online first,” Arvind et al² go so far as to suggest that their observation of this combination is “unique.” This seems somewhat of an exaggeration since in the text they cite the earlier report.¹ This report itself acknowledged the previous account provided by Alhadlaq et al,³ which revealed the presence of at least 11 known cases. The careful analysis provided by the latter authors is the more significant since they also listed several additional instances in which anomalies had been observed of the left coronary artery in the setting of an aortopulmonary window. As Márquez et al also commented in their report “Multiple reports of the association of aortopulmonary window and coronary arteries anomalies are published, but most of them are with an anomalous right coronary artery.” Does this not indicate that the association between the window and coronary arterial anomalies is far from rare? We have already suggested that the association might be more than coincidental, based on our finding of both lesions in autopsied hearts.⁴ It is unlikely that any of these investigators would have encountered our initial account, since we were providing a review of our knowledge, at that time, of the development of the coronary arteries. When discussing development, nonetheless, Márquez et al opined that the window itself “is caused by incomplete fusion of the two opposing conotruncal ridges or incomplete development of the conotruncal septum, resulting in an abnormal communication distal to the semilunar valves.” To justify this statement, the authors cited an earlier review provided by the team working at the Herma Heart Center in Milwaukee.⁵ As we explained in our own review,⁴ the window is better explained on the basis of the failure of closure of the foramen found during development between the proximal edge of the aortopulmonary septum and the distal margin of the cushions that separate the middle and proximal parts of the developing outflow tract. Since publication of our initial review, we have continued our investigation of the development of the coronary arteries. Although still a “work in progress,” our findings show that, when first recognizable, the stems of the major coronary arteries originate from the developing intrapericardial aorta distal to the sinutubular junction.⁶ Only with ongoing development do the arteries achieve their definitive positions within the valvar sinuses. It is

entirely feasible, therefore, that persistence of the embryonic aortopulmonary foramen, resulting in a postnatal window, also disturbs the appropriate incorporation and formation of the coronary arteries. These more recent findings strengthen our suggestion that the combination of an aortopulmonary window with coronary arterial malformations should be anticipated, rather than considered “extremely rare.” It is certainly not unique.


Yours faithfully



Robert H. Anderson, BSc, MD, PhD (Hon),
FRCPATH, FRCS Ed (Hon) 

Biosciences Institute, Newcastle University, London, UK
Email: sejrjan@ucl.ac.uk

ORCID iD

Robert H. Anderson  <https://orcid.org/0000-0001-5163-9021>

References

- Márquez D, Chalela T, Sandoval N. Intraoperative diagnosis of ALCAPA complicating an aortopulmonary window repair. *World J Pediatr Congenit Heart Surg.* 2022;13(1):92-94. doi: 10.1177/2150135120975761
- Arvind B, Devagourou V, Saxena A. Unique association of aortopulmonary window with anomalous origin of left coronary artery from pulmonary artery in an infant: a blessing in disguise? *World J Pediatr Congenit Heart Surg.* 2022;13(3):400-403. doi: 10.1177/21501351211060033
- Alhadlaq A, Dhillon S, Hancock-Friesen CL, Hussain A. A hidden culprit for ventricular dysfunction in aortopulmonary window repair: anomalous origin of left coronary artery. Case report and review of the literature. *J Thorac Cardiovasc Surg.* 2016;152(6):e123-e126.
- Spicer DE, Henderson DJ, Chaudhry B, Mohun TJ, Anderson RH. The anatomy and development of normal and abnormal coronary arteries. *Cardiol Young.* 2015;25(8):1493-1503.
- Barnes M, Mitchell ME, Tweddell JS. Aortopulmonary window. *Semin Thorac Cardiovasc Surg Pediatr Card Surg Ann.* 2011; 14(1):67-74.
- Anderson RH, Turner JE, Henderson DJ. The morphogenesis of abnormal coronary arteries in the congenitally malformed heart. *J Thorac Cardiovasc Surg.* 2021; S0022-5223(21)01406-9. Online ahead of print. doi: 10.1016/j.jtcvs.2021.08.084

Response to Comment on Aortopulmonary Window and Anomalies of Coronary Arterial Origin

Submitted February 12, 2022; Accepted February 19, 2022

We thank Professor Anderson for his comments on our paper wherein we discussed the challenges faced during the management of a patient

with anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) associated with aortopulmonary window (APW).¹ As acknowledged in our report, a handful of cases with a similar combination are available in literature. In addition, Prof Anderson proposes, based on his relentless observations, that there might be an embryologic plausibility for the occurrence of coronary arterial malformation in hearts with persistent aortopulmonary foramen. Hence, abnormalities of coronary origin may be anticipated in patients with APW. We do concur with him that this combination might not be as “unique” as we had claimed to be in our title, but still is quite