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Coarctation of the aorta associated with anomalous origins of the coronary arteries

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Coarctation of the aorta (CoA) is a complex disease of the vasculature with prevalence around 0.6% to 0.8% in general population and is even one hundred times higher in patients with congenital heart disease [1]. It can be treated by surgical or percutaneous (balloon and/or stent) methods. Of note, any coexisting cardiovascular malformations could hamper such interventions and might require targeted corrections at the same or staged procedure.

Anomalous coronary arteries (ACA) represent a heterogeneous group. Anomalous aortic origin of coronary arteries (AAOCA) is a rare finding in invasive angiography or computed tomography angiography (CTA) examinations (around 1.5%) [2]. Significantly, and similar to CoA, ACA may coexist with other cardiovascular malformations. There are some reports of the coexistence of CoA and anomalous coronary artery from pulmonary artery (ACAPA) (Table 1). However, there is paucity of data on the prevalence and clinical relevance of coexisting CoA and AAOCA.

Herein is retrospectively described, the clinical and imaging data of patients with a history of CoA and coexisting ACA. Methods: between January 2008 and November 2020 individual patient's discharge diagnoses and electronic records of chest CTA and invasive coronary angiography were retrospectively screened for the presence of AAOCA and ACAPA among consecutive patients, who were hospitalized at the documented single-center.

In the studied period 103,330 patients were hospitalized. Among them 372 subjects with a diagnosis of CoA were identified (prevalence of CoA in patients hospitalized at our institution – 0.36%). Among these CoA-patients, AAOCA was present in 5 subjects (1.3%, 5/372). Thus, the frequency of CoA-patients with AAOCA among all hospitalized patients was 0.005% (5/103,330). Patients with CoA and ACAPA were not identified. Demographics, clinical data and interventions are shown in Supplementary Table 2. Three patients had arterial hypertension, and one patient had atrial fibrillation.

All these five patients underwent ECG-gated CTA of the aorta (before or after corrective intervention).

One patient had simple coexistence of CoA with ACA, whereas 4 patients had other concomitant heart defects (including two patients complex cardiovascular defects).

RCA originating from the left sinus of Valsalva (SoV) was present in 2 patients, left circumflex

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Table 1. Brief literature review of the coexistence of the anomalous coronary artery and the coarctation of the aorta (by number of patients, leading message and chronologically)

Author	Number of patients/age at the diagnosis	Leading message	Type of ACA
I. CoA and ACA originating from the aorta (AAOCA)			
Children			
Awasthy et al. 2015	1/13 d	Surgical repair of CoA and closure of fistula	LAD fistula to RV
Kellogg et al. 1934	1/16 y	Coexistence of CoA, ACA and PDA. Postmortem finding.	Bifid mouth of RCA
Adults			
Tuzcu et al. 1990	6/ns	Angiographic study	Not specified
Ganga et al. 2018	1/54 y	First report of CoA and LCANCS	LCA from non-coronary SoV
Ayusawa et al. 2010	1/19 y	Coexistence of CoA, ACA and BAV	RCA from L SoV
Topaz et al. 1992	1/ns	Coexistence of CoA, ACA and VSD	Anomalous LCx
II. CoA and ACA originating from the pulmonary artery (ACAPA)			
Neonates/children			
Laux et al. 2014	5/ns	Preoperatively missed ALCAPA in 4 cases	ALCAPA
Levin et al. 1990	2/7.5 w and 7.5 m	First report	ALCAPA
Santoro et al. 1995	2/1 m and 2 m	Preoperatively missed ACAPA	SCA from PA
Morgan et al. 2010	2/5 m and 8 m	Coexistence of ALCAPA, CoA and MS	ALCAPA
Hölscher et al. 2019	1/18 d	Preoperative assessment	ALCAPA
Soni et al. 2019	1/7 y	Interrupted aortic arch	RCA from PA
Stefek et al. 2019	1/7 d	Aortic arch hypoplasia	RCA from PA
Liang et al. 2016	1/3 m	Congestive HF	ALCAPA
Aktas et al. 2015	1/15 m	Preoperatively missed ALCAPA	LCx from RPA
Illic et al. 2014	1/8 d	Coexistence of ACAPA, CoA, Scimitar syndrome	ALCAPA
Sekelyk et al. 2014	1/2 m	Preoperative assessment	LCx from RPA
Vitanova et al. 2014	1/newborn	Preoperatively missed ALCAPA	ALCAPA
Celik et al. 2010	1/10 d	Preoperatively missed ALCAPA. Coexistence of ALCAPA, CoA, CS orifice atresia and left SVC	ALCAPA
Radha et al. 2004	1/39 d	Preoperatively missed ALCAPA	ALCAPA
Moideen et al. 2004	1/39 d	Preoperatively missed ALCAPA	ALCAPA
Clarke et al. 2003	1/15 y	Preoperatively missed ALCAPA, Coexistence ALCAPA, CoA and BAV	LAD from PA
McMahon et al. 2002	1/infant	Interrupted aortic arch	ALCAPA
Da Cruz et al. 1998	1/12 d	Coexistence of ALCAPA, CoA, ASD, VSD	ALCAPA
		Preoperatively missed ALCAPA	
Murakami et al. 1997	1/2 y	Preoperatively missed ALCAPA Re-do in adulthood	ALCAPA
Sarioglu et al. 1997	1/4 y	Preoperatively missed anomalous LCx	LCx from RPA
		Re-do for subaortic membrane and anomalous LCx	
Ma et al. 1994	1/1 d	Interrupted aortic trunk	ALCAPA

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Table 1. cont. Brief literature review of the coexistence of the anomalous coronary artery and the coarctation of the aorta (by number of patients, leading message and chronologically)

Author	Number of patients/age at the diagnosis	Leading message	Type of ACA
Tsuchiya et al. 1989	1/2 m	Coexistence of ALCAPA, CoA, AS, PDA	LAD from PA
Bharati et al. 1984	1/3.5 m	Postinterventional death	ALCAPA
Driscoll et al. 1982	1/3 w	Coexistence of ALCAPA and complex heart defect	ALCAPA
Honey et al. 1975	1/13 y	Coexistence of ALCAPA, CoA, RAA	LCx from RPA
Alexander et al. 1956	1/5 m	Coexistence of ALCAPA, CoA, PDA Autopsy study	ALCAPA

ACA — anomalous coronary anomaly, ACAPA — anomalous coronary artery from the pulmonary artery, ALCAPA — anomalous left coronary artery from the pulmonary artery, ALVT — aorto-left ventricular tunnel, AS — aortic stenosis, ASD — atrial septal defect, BAV — bicuspid aortic valve, CoA — coarctation of the aorta, CS — coronary sinus, CTA — computed tomography angiography, d- days, HF — heart failure, KD — Kawasaki disease, L — left, LAD- left anterior descending coronary artery, LCA — left coronary artery, LCANCS — left main coronary artery from noncoronary sinus, LCx — left circumflex coronary artery, LV — left ventricle, m- months, MS — mitral stenosis, ns- not specified, OI — osteogenesis imperfecta, PA — pulmonary artery, PDA — patent ductus arteriosus, R — right, RAA — right aortic arch, RCA — right coronary artery, RPA — right pulmonary artery, RV — right ventricle, SCA — single coronary artery, SCD — sudden cardiac death, SoV — sinus of Valsalva, SVC — superior vena cava, TAV — tricuspid aortic valve, VSD — ventricular septal defect, w — weeks, y — years

coronary artery originating from the right SoV was present in 1 patient, left coronary artery originating above the left SoV was detected in 1 patient, whereas RCA originated from non-coronary cusp was visualized in 1 patient.

Bicuspid aortic valve was visualized in 1 patient, whereas another patient had significant aortic stenosis. No myocardial bridging of the coronaries was identified by CTA.

All 5 patients underwent prior surgical correction of CoA (4 of them in the adulthood). One patient had isolated operation of CoA, whereas 2 patients had combined operation of CoA with patent ductus arteriosus or aortic valve replacement. Two patients underwent staged surgeries of concomitant cardiac malformations. One patient was treated percutaneously with stent implantation in the aortic arch at follow-up. None of the AAOCA were corrected.

This study presents a small series of CoA-patients with coexisting AAOCA. Heterogeneity of this group is expressed by patient's age at the time of first CoA intervention (range from 1 month to 57 years), type of AAOCA and concomitant heart defects.

To date, few and mostly single reports described a combination of CoA with ACA – the largest series included 6 patients with no details of ACA types [3]. As mentioned before, ACAPA or more specifically anomalous left coronary artery from the pulmonary artery (ALCAPA) were reported more frequently than AAOCA in CoA-patients (Table 1) [4]. This is opposite to the present observations, where ACAPA was not identified among the CoA cohort. The possible explanation is that previous descriptions included cases of ACAPA and CoA almost exclusively in the pediatric population, whilst in the present group there was only one child (1-month old infant). Further, the prevalence of AAOCA among the CoA-patients (1.3%) is very similar to the reported prevalence of AAOCA in the general population (1.5%) [2].

AAOCA may extend from benign types (although they may pose difficulties during invasive coronary angiography — e.g. high coronary take-off) to potentially life-threatening AAOCA with a malignant course (such as LAD traversing between aorta and pulmonary trunk with its significant compression). The latter type of AAOCA was not observed in the present series and thus, none of the patients underwent surgical correction of AAOCA. The frequency of specific types of AAOCA found in the current cohort was pretty much the same as in previous studies of AAOCA by invasive angiography [5] and CTA [6].

From a practical point of view, surgical correction of CoA requires the left lateral thoracotomy approach. Of note, any correction of heart defects (including malignant AAOCA) may not be possible through this approach [7]. A decision to operate on the AAOCA during surgical correction of CoA may be controversial in some cases. Recommendations for surgical management of AAOCA are covered in detail by the expert consensus guidelines [8]. Briefly, inter-arterial AAOCA course is a strong recommendation for surgical reconstruction for

symptomatic patients and (with lesser level of evidence) for asymptomatic patients with anomalous inter-arterial course of the left coronary artery. Management of risk stratification of asymptomatic patients with anomalous RCA originating from the opposite SoV is uncertain. Thus, indication for surgical treatment remains a matter of discussion and may be individualized [9]. There are different types of surgical repair of AAOCA. Of note, the only possible coronary correction through the left thoracotomy (used for surgical management of CoA) is the coronary bypass to the left anterior descending coronary artery (LAD) using left internal mammary artery.

The current study was not free of limitations. First, it had a retrospective design. Second, a heterogenous patient population was included based on pre-specified search criteria rather than meticulous analysis of all consecutive CT scans. Finally, the presented prevalence of CoA and AOOCA represents data from patients who were hospitalized in a single cardiac center and not from the general population.

Coexistence of CoA and AAOCA is very rare and seems to be purely incidental. Detailed screening for AAOCA during preinterventional work-up of patients with congenital heart disease (including CoA) is advisable, as it may have potential implications during corrective interventions.

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