ARCHIVES OF THE TURKISH SOCIETY OF CARDIOLOGY

Official journal of the TURKISH SOCIETY OF CARDIOLOGY

Anomalous Origin of the Left Coronary Artery from the Pulmonary Artery Syndrome in Adults: A Case-Based Clinical Review

Yetişkinlerde Pulmoner Arterden Anormal Olarak Kaynaklanan Sol Koroner Arter Sendromu: Vaka Temelli Klinik İnceleme

ABSTRACT

This review provides a structured overview of ten published case reports of anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) syndrome in adults, with a focus on clinical presentation, diagnostic methods, and treatment approaches. A narrative review was conducted using the PubMed database to identify English-language case reports of adult patients with ALCAPA. Ten cases were selected based on clearly reported clinical features, diagnostic methods, and treatment approach. The most common symptoms included atrial fibrillation, chest pain, dyspnea, and syncope. Surgical correction was performed in four cases, while others were managed conservatively or with medical therapy. Adult-type ALCAPA presents with variable clinical manifestations and requires individualized treatment. Timely diagnosis and appropriate management are crucial for optimal outcomes.

Keywords: ALCAPA syndrome, congenital heart diseases, sudden cardiac death

ÖZET

Bu derleme, yetişkinlerde pulmoner arterden sol koroner arterin anormal kökenli (ALCAPA) sendromuna ilişkin yayınlanmış on vaka raporunun klinik görünüm, tanı yöntemleri ve tedavi yaklaşımlarına odaklanarak yapılandırılmış bir özetini sunmaktadır. ALCAPA'lı yetişkin hastaların İngilizce vaka raporlarını belirlemek için PubMed veritabanı kullanılarak anlatımsal bir derleme yapılmıştır. Açıkça bildirilen klinik özellikler, tanı yöntemleri ve tedavi yaklaşımına göre on vaka seçilmiştir. En sık görülen semptomlar atriyal fibrilasyon, göğüs ağrısı, dispne ve senkop idi. Dört vakada cerrahi düzeltme yapılırken, diğer vakalar konservatif veya tıbbi tedavi ile yönetilmiştir. Yetişkin tipi ALCAPA, değişken klinik belirtilerle ortaya çıkar ve bireyselleştirilmiş tedavi gerektirir. Optimal sonuçlar için zamanında tanı ve uygun yönetim çok önemlidir.

Anahtar Kelimeler: ALCAPA sendromu, konjenital kalp hastalıkları, ani kardiyak ölüm

REVIEW
DERLEME

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Received: July 03, 2025
Accepted: September 23, 2025

Cite this article as: Spiljak A. Anomalous Origin of the Left Coronary Artery from the Pulmonary Artery Syndrome in Adults: A Case-Based Clinical Review. *Turk Kardiyol Dern Ars.* 2025;53(0):000-000.

DOI: 10.5543/tkda.2025.98044

Anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA), also known as Bland–White–Garland syndrome, is a rare but potentially life-threatening congenital coronary anomaly. Without timely surgical correction, the condition is associated with high morbidity and mortality, particularly in infancy. ALCAPA is classified into two main types based on the age and nature of presentation: the infant type, which typically manifests within the first few months of life with signs of myocardial ischemia or heart failure, and the adult type, which is less common and may remain undiagnosed until later in life.[1] ALCAPA most commonly presents as an isolated congenital coronary anomaly, although it has occasionally been associated with other structural defects, including tetralogy of Fallot, coarctation of the aorta, ventricular septal defect, and patent ductus arteriosus. The anomaly remains clinically silent during fetal life due to comparable pressures and oxygenation in the great arteries. However, after birth, the progressive decline in pulmonary artery pressure leads to insufficient coronary perfusion, resulting in myocardial ischemia. This cascade may cause left ventricular dysfunction, chamber dilation, and mitral regurgitation, ultimately manifesting as heart failure, arrhythmias, or sudden cardiac death in the absence of timely recognition and

treatment.[2,3] Echocardiography remains the most commonly used initial diagnostic tool for ALCAPA. When findings such as left ventricular dilation and dysfunction, significant mitral regurgitation, abnormal flow patterns across the interventricular septum, and right coronary artery dilatation are present, along with increased echogenicity and thickening of the endocardium, clinicians should extend their differential diagnosis beyond idiopathic endocardial fibroelastosis (EFE) and idiopathic dilated cardiomyopathy (DCM). Careful assessment of the coronary artery origins is crucial to avoid misdiagnosis. The hallmark echocardiographic sign of ALCAPA is visualization of the left coronary artery arising from the pulmonary artery, with delineation of its origin, size, and course. The presence of reversed flow within the left coronary artery and a well-developed collateral network further support the diagnosis. Owing to its noninvasive nature, repeatability, affordability, and ability to visualize coronary origins, echocardiography is considered indispensable. It enables real-time assessment of hemodynamic status, early detection of subclinical myocardial dysfunction, and reliable postoperative follow-up through evaluation of cardiac size, wall motion, contractility, and strain rate. Nevertheless, digital subtraction angiography (DSA) with coronary angiography (CAG) remains the gold standard for confirming the diagnosis of ALCAPA. In neonates and infants, however, the use of contrast agents is sometimes controversial due to concerns about radiation exposure and potential allergic reactions. Noninvasive modalities such as coronary computed tomography angiography (CCTA) and magnetic resonance angiography (MRA) are increasingly employed. CCTA offers excellent spatial resolution and is particularly useful for visualizing small vessels like the coronaries, although it does not provide information on intravascular flow. MRA, on the other hand, is useful in evaluating postoperative ventricular morphology and function, particularly in suspected myocardial ischemia, although image quality may be affected by elevated heart rates.[4] The 2018 guidelines from the American Heart Association (AHA) and American College of Cardiology (ACC) recommend surgical correction of ALCAPA in all patients, regardless of age or symptoms, due to the lifelong risk of myocardial ischemia, ventricular arrhythmias, and sudden cardiac death. Surgical techniques are generally categorized into single- and dual-coronary system repairs. In the past, simple ligation of the anomalous artery—a single-coronary approach—was common, but today it is used only temporarily in critically ill infants to prevent coronary steal and allow clinical stabilization prior to definitive repair. The current surgical objective is to restore a dual coronary circulation. Techniques include direct reimplantation of the anomalous artery into the aorta (coronary button transfer), the Takeuchi procedure (intrapulmonary baffle), subclavian-to-left coronary artery anastomosis, and coronary artery bypass grafting (CABG) using venous or arterial grafts. Cardiac transplantation is reserved for patients with severe left ventricular dysfunction and refractory heart failure. The coronary button technique is preferred in infants because it offers near-physiologic perfusion with low complication rates. In adult patients, direct reimplantation may not be feasible due to the immobility or fragility of the anomalous artery, so the Takeuchi technique or CABG is typically performed. Although surgery remains the standard treatment, medications may be used adjunctively—for example, in elderly patients with high surgical risk, or in infants as preoperative support to improve cardiac function.[5]

ABBREVIATIONS

ACC	American College of Cardiology
ACEI	Angiotensin-converting enzyme inhibitors
AF	Atrial fibrillation
AHA	American Heart Association
ALCAPA	Anomalous origin of the left coronary artery from the pulmonary artery
ARBs	Angiotensin II receptor blocker
CABG	Coronary artery bypass grafting
CAG	Coronary angiography
CCTA	Coronary computed tomography angiography
CMR	Cardiac magnetic resonance
CXR	Chest radiography
DCM	Idiopathic dilated cardiomyopathy
DSA	Digital subtraction angiography
ECG	Electrocardiography
EFE	Idiopathic endocardial fibroelastosis
ICD	Implantable cardioverter-defibrillator
IMA	Internal mammary artery
LAD	Left anterior descending artery
LAVI	Left atrial volume index
LCA	Left coronary artery
LCx	Left circumflex artery
LMCA	Left main coronary artery
LV	Left ventricle
LVEF	Left ventricular ejection fraction
MDCT	Multi-detector computed tomography
MPI	Stress myocardial perfusion imaging
MR	Mitral regurgitation
MRA	Magnetic resonance angiography
PA	Pulmonary artery
PCI	Percutaneous coronary intervention
RCA	Right coronary artery
ROSC	Return of spontaneous circulation
SVG	Saphenous vein graft

Materials and Methods

A systematic search of the PubMed database was performed in June 2025. The search was restricted to case reports published in English between January 2020 and June 2025, involving adult patients (> 18 years). Pediatric and borderline cases were excluded. The following search terms were used: "ALCAPA syndrome adult" (yielding 19 records) and "Bland-White-Garland syndrome adult" (yielding 14 records). After removal of duplicates (n = 20) and exclusion based on title/abstract (n = 3), ten full-text case reports were assessed for eligibility. Only full-text, freely accessible articles were included. The selection process followed the PRISMA 2020 guidelines (Preferred Reporting Items for Systematic reviews and Meta-Analyses) and is summarized in the corresponding flow diagram (Figure 1). Additional case reports were identified but excluded from the final analysis due to incomplete data or non-English full text.

Results

Ten case reports of adult patients diagnosed with anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) were included in the analysis. Patient age ranged from 29 to 81 years, and the majority were female (9 out of 10).

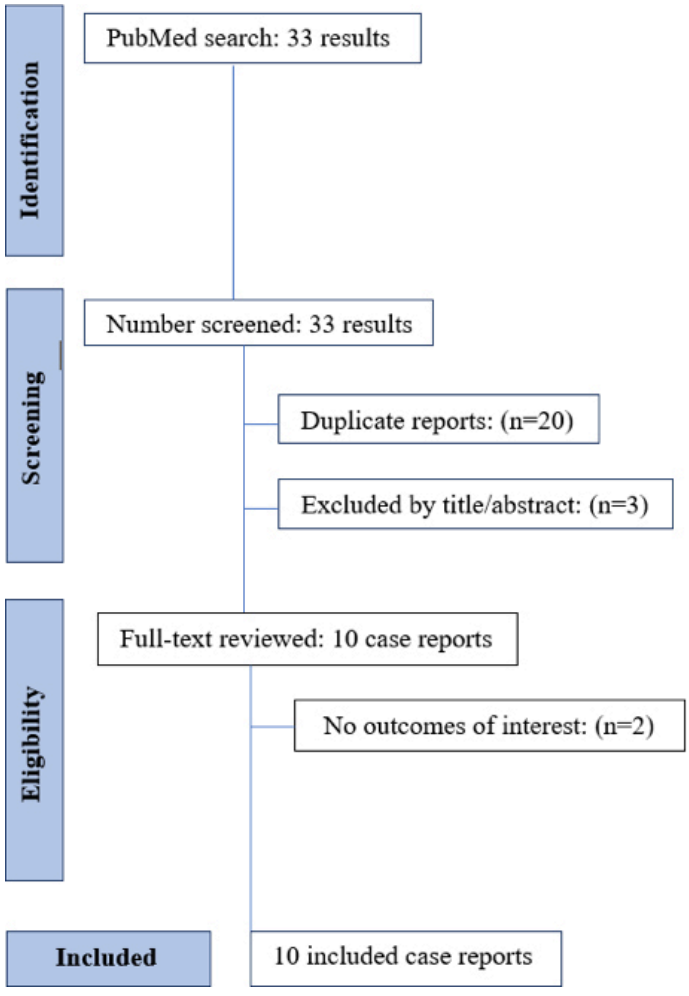

Figure 1. PRISMA flow diagram of study screening and selection.

The extracted data included age, sex, clinical presentation, diagnostic modalities, and treatment strategies. An overview of the key findings is presented in Table 1.[6-15]

The most commonly reported presenting symptoms included atrial fibrillation, chest pain, dyspnea, and syncope. However, a subset of patients remained asymptomatic at the time of diagnosis, with the condition detected incidentally during preoperative assessment or workup for unrelated complaints (Table 1). This highlights the potential for ALCAPA to remain clinically silent until late adulthood. Notably, three of the included cases exhibited acute and severe clinical presentations, namely acute myocardial infarction, sudden cardiac arrest, and acute heart failure, underscoring the potential of ALCAPA to present as a life-threatening emergency in adult patients (Figure 2).

In nearly all cases, transthoracic or transesophageal echocardiography, along with coronary imaging—either computed tomography angiography (CTA) or catheter-based coronary angiography (CAG)—played a central role in establishing the diagnosis. Additional modalities such as cardiac magnetic resonance imaging (MRI) and stress myocardial perfusion imaging were employed in select cases to assess myocardial viability, presence of ischemia, and the extent of collateral circulation (Figure 3, 4).

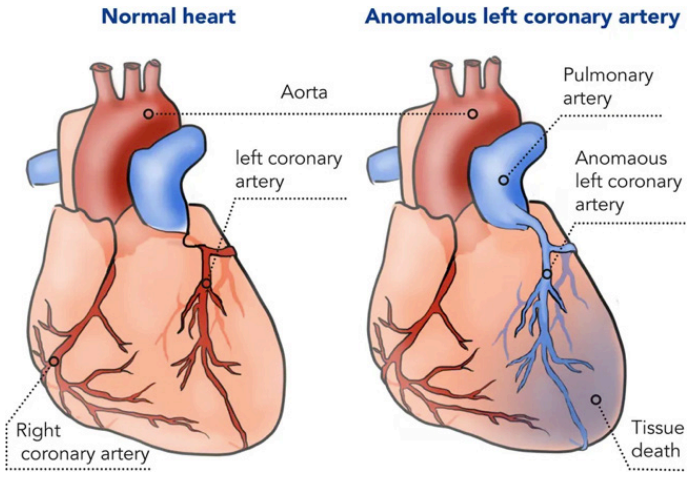

Figure 2. Schematic illustration of a normal heart and anomalous left coronary artery from the pulmonary artery (ALCAPA). Reproduced with permission under the Creative Commons Attribution-NonCommercial-NoDerivatives 4.0 International License from Kyorin University.

Therapeutic strategies varied considerably across cases (Table 1). Four patients underwent surgical repair, most commonly direct reimplantation of the anomalous left coronary artery into the aorta, sometimes combined with grafts or concomitant valve procedures, with favorable outcomes and resolution of symptoms. Another four patients were managed conservatively, either due to adequate collateralization, absence of ischemia on stress imaging, or refusal of surgery, and all remained clinically stable on follow-up. One patient underwent percutaneous coronary intervention (PCI) with angioplasty of the left circumflex artery and was discharged in good condition. One patient died before definitive therapy could be performed.

Follow-up durations ranged from one month to three years, with generally favorable outcomes in both surgically and conservatively managed patients.

Where reported, postoperative functional outcomes demonstrated significant improvement in left ventricular function and symptom relief, whereas NYHA (New York Heart Association) classification was inconsistently documented across reports.

These findings underscore the clinical heterogeneity and diagnostic challenges of adult-type ALCAPA. Management must be tailored individually, based not only on symptom burden but also on myocardial viability, coronary anatomy, surgical risk, and patient preferences. Given the potential for sudden cardiac events even in minimally symptomatic individuals, early recognition and appropriate risk stratification remain crucial.

Discussion

Anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) is an extremely rare congenital anomaly in adults, typically diagnosed during evaluation for arrhythmia, ischemic symptoms, or incidentally. In ALCAPA, the left coronary artery originates abnormally from the pulmonary artery, resulting in perfusion of the left ventricle with desaturated blood at low pressure. Given the high oxygen demand of the left

Table 1. Summary of adult anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) case reports

Author (year)	Age (years)	Sex	Clinical presentation	Diagnosis	Treatment	Follow-up duration	Long-term outcomes	NYHA functional class
Lotman et al. (2020)[6]	76	F	Asymptomatic at presentation; history of palpitations and irregular pulse (AF detected)	ECG, echocardiography, CAG, CCTA	Conservative management	3 years	Good physical condition, no adverse events	Not reported
De Stefano et al. (2024)[7]	76	F	Fever, fatigue, cognitive-motor slowing, hyposthenia of the left hemisoma	ECG, echocardiography, CXR, cranial computed tomography, chest CT, contrast-enhanced CT with ECG	Fatal outcome prior to surgical or medical management	1 month	Death (January 2022)	Not reported
Niu et al. (2022)[8]	64	F	Paroxysmal precordial pain for 10 years, aggravated over 2 days, acute myocardial infarction	ECG, echocardiography, CCTA, aortic angiography, MDCT	Surgical correction recommended but refused; medical therapy initiated	6 months	No significant chest pain during follow-up	Not reported
Prandi et al. (2022)[9]	55	F	Syncope, ventricular fibrillation, sudden cardiac arrest, AF after ROSC	ECG, echocardiography, 24-hour ECG Holter monitoring, head CT, CCTA, cardiac catheterization, MPI, CMR, MRA	Surgical repair with direct LCA reimplantation using Hemashield grafts; ICD placement; medical therapy	3 months	Asymptomatic, no surgical complications, returned to normal activity	Not reported
Rao et al. (2022)[10]	29	F	Dyspnea, orthopnea, edema, weight gain; AF, heart failure	ECG, echocardiography, CXR, cardiac catheterization, CCTA	Conservative management	Not specified (on regular follow-up after discharge)	Stable on medical therapy, maintained sinus rhythm post-cardioversion, no urgent surgery required	Not reported
Alsamman et al. (2021)[11]	38	F	Chest pain, palpitations, diaphoresis during exercise, AF	ECG, echocardiography, CAG, cardiac catheterization, CCTA, head CT	LMCA translocation to aorta; full recovery	Short-term and ongoing tertiary center follow-up	Recovered well, no complications	Not reported
Riaz Gondal et al. (2024)[12]	81	F	Chest pain, progressive mental deterioration, bradycardia, hypotension, complete heart block	ECG, cardiac catheterization	Atropine, transcutaneous and temporary transvenous pacing, percutaneous balloon angioplasty of LCx; discharged with ACS medical therapy	Short-term follow-up until discharge	Discharged stable, no pacing needed after PCI	Not reported
Sandugash Talkhatova et al. (2023).[13]	52	F	Asymptomatic (incidentally detected during routine check-up)	ECG, echocardiography, CAG, CCTA, cardiac catheterization	Conservative management	3 months; planned follow-up every 6 months	Asymptomatic at 3 months, good exercise tolerance, no adverse events	Not reported
Tetsuya Saito et al. (2023).[14]	65	F	Palpitations, mild dyspnea, AF	ECG, echocardiography, CXR, CCTA	Surgical repair (ALCAPA repair, mitral valve repair, Cox-Maze IV, cryoablation, CABG with SVG to LAD)	1 year (regular follow-up described)	Good condition, sinus rhythm, improved LVEF (70%), reduced LAVI, mild MR, no AF recurrence	Not reported
Maaweya Jabareen et al. (2024).[15]	27	M	Shortness of breath that had gradually worsened over the past few months	ECG, echocardiography, CCTA, cardiac catheterization	Conservative management	2 years	Good condition, asymptomatic, no reported adverse events	Not reported

ECG, Electrocardiography; CAG, Coronary angiography; CCTA, Coronary computed tomography angiography; CXR, Chest radiography; CT, Computed tomography; MPI, Myocardial perfusion imaging; CMR, Cardiac magnetic resonance; MRA, Magnetic resonance angiography; MDCT, Multi-detector computed tomography; LCA, Left coronary artery; ICD, Implantable cardioverter-defibrillator; PCI, Percutaneous coronary intervention.

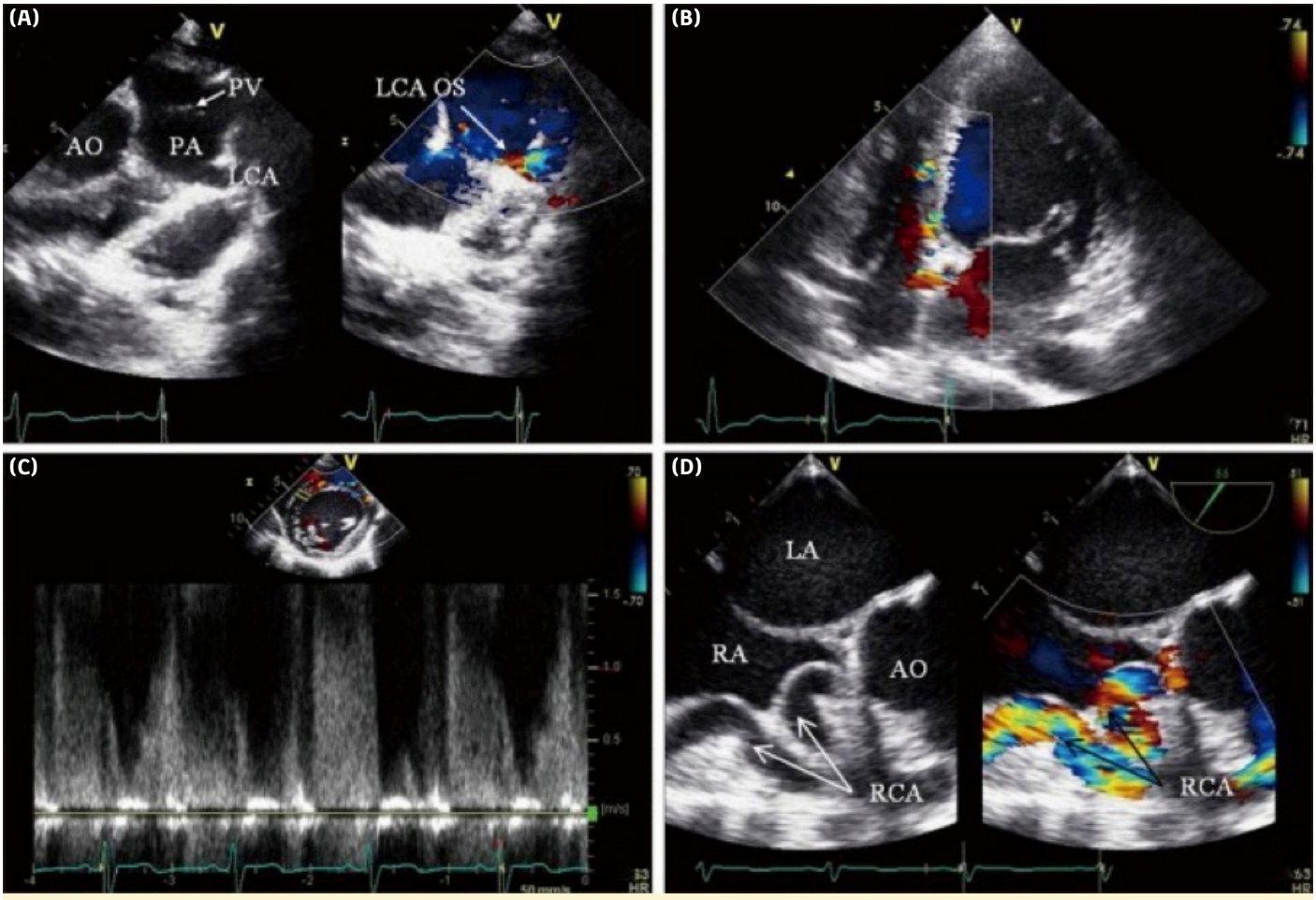

Figure 3. Transthoracic and transesophageal echocardiographic findings in a patient with anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA). (A) Left coronary artery (LCA) arising from the main pulmonary artery (PA); (B) color Doppler showing abundant septal collateral flow; (C) retrograde filling of the left anterior descending artery (LAD) on Doppler; (D) markedly dilated and tortuous right coronary artery (RCA). Reproduced from Kim et al., J Cardiovasc Ultrasound, 2012, under the Creative Commons Attribution Non-Commercial License (CC BY-NC 3.0).

ventricle, this leads to chronic myocardial ischemia, particularly during exertion. As pulmonary vascular resistance falls after birth, blood flow from the left coronary artery is diverted into the pulmonary artery, a process known as the coronary steal phenomenon, which further aggravates ischemia. Collateral circulation from the right coronary artery to the left coronary artery may partially compensate and allow survival into adulthood, but it rarely provides adequate myocardial perfusion. Consequently, progressive myocardial damage, infarction of the anterolateral left ventricular wall, and congestive heart failure may develop. Even in patients with extensive collaterals, the condition carries a risk of serious complications, including arrhythmias, angina, syncope, and sudden cardiac death. The clinical variability in adult patients with ALCAPA largely depends on the extent and adequacy of collateral circulation between the right and left coronary arteries. Patients with well-developed collaterals may maintain sufficient myocardial perfusion to remain asymptomatic, sometimes for decades. In contrast, when collaterals are insufficient, myocardial ischemia develops, leading to angina, arrhythmias, heart failure, or even sudden cardiac arrest. Additional factors such as left ventricular remodeling, the severity of the coronary steal phenomenon, and the presence of associated valvular dysfunction (e.g., mitral regurgitation secondary to papillary muscle ischemia) further contribute to symptom development. This pathophysiological spectrum explains why some adults are incidentally diagnosed during routine evaluation, while others present with life-threatening complications.[16]

While conventional coronary angiography with digital subtraction angiography has historically been regarded as the gold standard for diagnosing ALCAPA, its invasive nature and associated risks (ionizing radiation, contrast reactions) limit its use, particularly in infants. In contrast, modern multimodal imaging techniques provide complementary strengths (Figure 5):

1. CCTA offers high spatial resolution and direct visualization of the anomalous origin and collateral circulation. Its sensitivity for anatomic definition makes it invaluable for preoperative planning, though it cannot evaluate dynamic intravascular flow.
2. MRA provides functional assessment, including left ventricular morphology, myocardial ischemia, and viability,

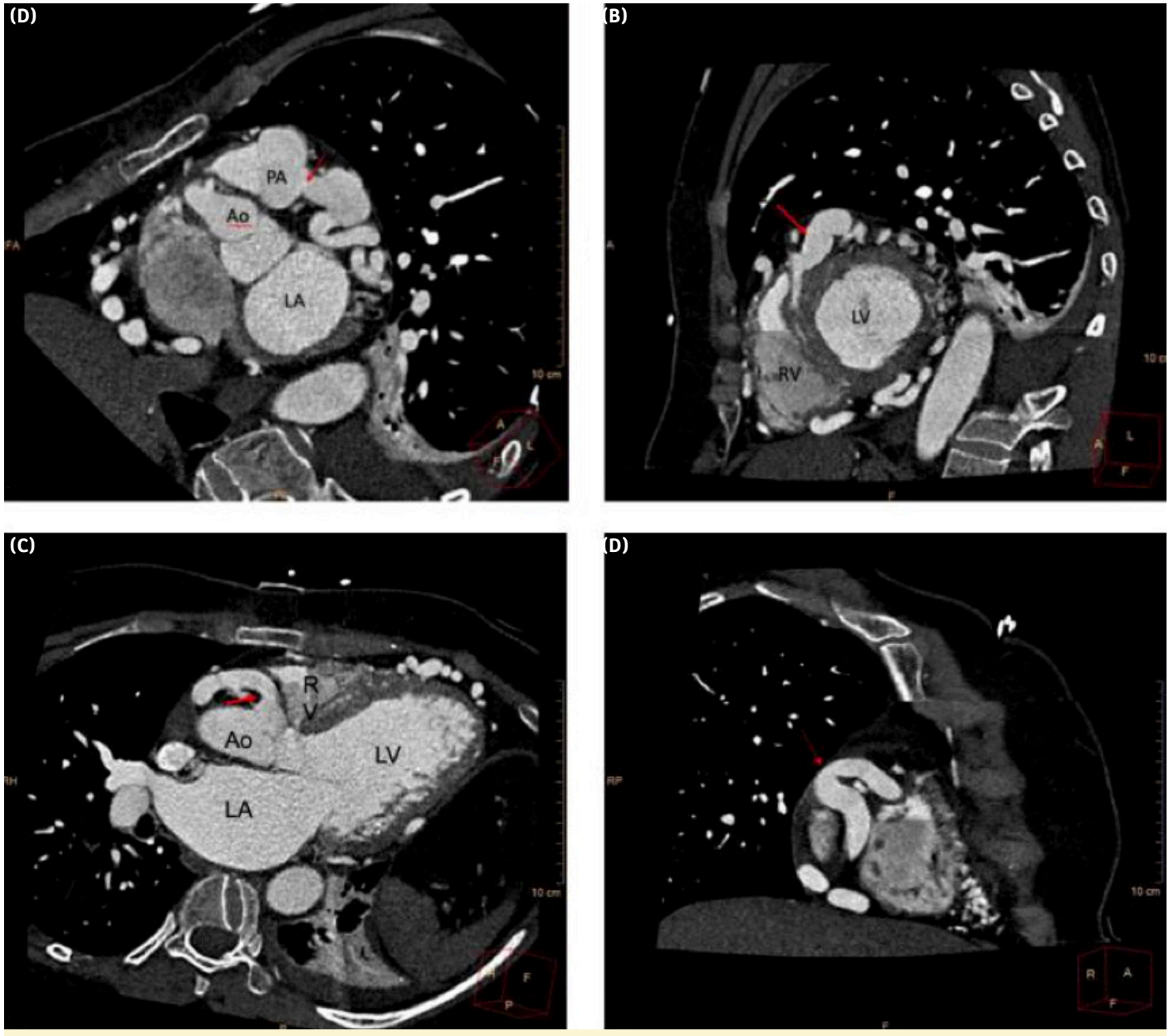

Figure 4. Coronary computed tomography angiography (CCTA) showing: (A) anomalous origin of the left coronary artery (red arrow) from the pulmonary artery (PA); (B) ectatic and tortuous left coronary artery (LCA) (red arrow); (C) normal origin of the right coronary artery (RCA, red arrow) from the right coronary sinus; and (D) diffusely ectatic coronary vasculature. Reproduced with permission from Prandi FR, Zaidi AN, LaRocca G, et al. Sudden Cardiac Arrest in an Adult with Anomalous Origin of the Left Coronary Artery from the Pulmonary Artery (ALCAPA): Case Report. Int J Environ Res Public Health. 2022;(3):1554. Licensed under Creative Commons Attribution (CC BY) 4.0 International License.

without radiation exposure. However, image quality can be affected by patient motion and high heart rates.

3. Conventional angiography remains superior in evaluating coronary steal physiology and offers the possibility of interventional treatment.

Therefore, combining modalities maximizes diagnostic accuracy: CCTA for structural definition, MRA for functional and viability data, and CAG for hemodynamic evaluation and therapeutic decision-making. This integrated approach improves both preoperative assessment and postoperative follow-up.[17]

The 2018 American Heart Association and American College of Cardiology guidelines recommend surgical correction of ALCAPA in all patients, regardless of age or symptoms, due to the lifelong risk of myocardial ischemia, malignant arrhythmias, and sudden cardiac death. The overarching goal of surgery is to re-establish a dual coronary system and ensure adequate myocardial perfusion. Surgical approaches include:

- Direct reimplantation of the left coronary artery (LCA) into the aorta (coronary button transfer): the gold standard, restoring near-physiologic circulation with excellent long-term outcomes.

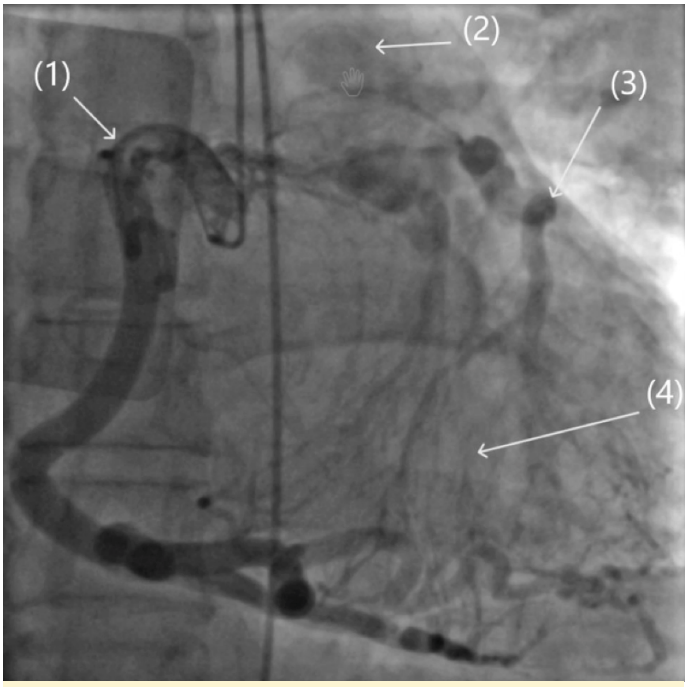

Figure 5. Coronary angiography demonstrating a markedly dilated right coronary artery (1), extensive collateral circulation (4), retrograde filling of the left anterior descending artery (3), and contrast runoff into the pulmonary trunk (2). Reproduced from Urban T, Grundmann S, Klein F, Wengenmayer T, Müller-Peltzer K, Busch HJ. First diagnosis of ALCAPA syndrome in adulthood: a rare cause of cardiac arrest. Inn Med (Heidelb). 2025;66(1):124–128. German, under the terms of the Creative Commons Attribution (CC BY 4.0) license.

- Two-flap technique: a modification of reimplantation used when the LCA is too short to reach the aorta without tension.
- Takeuchi procedure (intrapulmonary baffle repair): creating an aortopulmonary window and baffle between the aorta and anomalous LCA; most common in pediatric patients but also used in adults.
- Coronary artery bypass grafting: using a saphenous vein graft (SVG) or internal mammary artery (IMA), generally reserved for older adults or when reimplantation is technically not feasible.

Adjunctive procedures include mitral valve repair for regurgitation, Cox-Maze IV or cryoablation for atrial fibrillation, and implantable cardioverter-defibrillator (ICD) implantation in patients with malignant arrhythmias. Cardiac transplantation is considered in patients with severe, irreversible left ventricular (LV) dysfunction and refractory heart failure.

Surgical repair is preferred in the vast majority of patients, including asymptomatic individuals, because of the lifelong risk of ischemia and sudden death. Younger and symptomatic patients (angina, arrhythmia, syncope, heart failure) almost invariably require surgery.

Conservative management may be considered in carefully selected adults with extensive collateral circulation, preserved LV function, no demonstrable ischemia on functional testing, or in those with prohibitive surgical risk (e.g., advanced age, significant comorbidities). In such cases, therapy typically includes beta-blockers, angiotensin-converting enzyme inhibitors (ACEIs), angiotensin II receptor blockers (ARBs), antiarrhythmic drugs, and anticoagulation/antiplatelet agents. Close clinical and imaging surveillance is mandatory.

Percutaneous coronary intervention has only a limited role, used primarily to treat coexisting obstructive coronary artery disease rather than the anomaly itself.

In summary, corrective surgery remains the gold standard, whereas conservative therapy is reserved for exceptional situations in which the risks of surgery outweigh the potential benefits.[5]

In the largest published review of 151 adult cases of ALCAPA, the mean age at diagnosis was 41 years, with the oldest patient being 83 years old. The majority (66%) presented with angina, dyspnea, palpitations, or fatigue, while 17% presented with malignant arrhythmias, syncope, or sudden cardiac death, and 14% were incidentally diagnosed. Notably, 12% of cases were identified only at autopsy, and most patients underwent surgical correction during their clinical course.[18]

In contrast, the present review of ten adult cases demonstrated an older age range (29–81 years, with a median above that reported in larger cohorts) and a predominance of female patients (9 out of 10). Clinical presentation was heterogeneous, ranging from incidental detection to life-threatening events such as acute myocardial infarction, cardiac arrest, or acute heart failure. While four patients underwent surgical repair and four were managed conservatively, one underwent percutaneous coronary intervention and one died before definitive treatment. These findings highlight that, despite the limited sample size, the spectrum of presentation and treatment strategies mirrors those of larger series, though with proportionally higher representation of conservatively managed patients.

Importantly, even after surgical repair, adult ALCAPA patients remain at risk for adverse events, including myocardial ischemia, arrhythmias, and sudden cardiac death, underscoring the need for long-term follow-up and cardiac surveillance.

Current literature on adult ALCAPA is limited to isolated case reports and small case series. Prospective data are lacking, and there are no standardized recommendations for long-term surveillance strategies in adult patients. Consequently, the choice between surgical and conservative management is largely individualized, depending on institutional expertise and patient-specific risk profiles. Future multicenter registries and prospective studies are needed to define optimal treatment algorithms and follow-up protocols for this rare but potentially lethal condition.

Clinical Implications

Early recognition of ALCAPA syndrome in adulthood remains crucial, as delayed diagnosis is associated with progressive myocardial ischemia, ventricular dysfunction, and a high risk of sudden cardiac death. Echocardiography, CCTA, and coronary angiography are the key diagnostic modalities that allow timely identification of the anomaly. Once diagnosed, surgical correction should be prioritized, as conservative management carries substantial risks and is rarely appropriate outside of

selected asymptomatic cases with preserved ventricular function. Given the lack of standardized long-term follow-up strategies, clinicians should remain vigilant in surveillance of postoperative patients. Future multicenter registries and prospective studies are needed to establish evidence-based recommendations for adult ALCAPA management.

Conclusion

ALCAPA syndrome is rare but serious, with variable presentation. Long-term follow-up and awareness are essential.

Conflict of Interest: The author have no conflicts of interest to declare.

Funding: The author declared that this study received no financial support.

Use of AI for Writing Assistance: AI-assisted technologies were not used in this article.

Peer-review: Externally peer-reviewed.

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