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Pediatric coronary cameral fistula in a structurally normal heart: a case report and review of the literature

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Abstract

Introduction Coronary cameral fistulas (CCFs) are rare congenital anomalies characterized by abnormal connections between a coronary artery and one of the cardiac chambers. These abnormal connections can lead to significant clinical implications, including heart failure and myocardial ischemia, necessitating timely diagnosis and intervention.

Case presentation A 5-year-old Iranian boy was brought to the emergency room at Heart Hospital Center complaining of chest pain and dyspnea on exertion. He had experienced similar episodes over the past 4 months, which had not been evaluated. Physical examination revealed stable vital signs and no remarkable findings. Transthoracic echocardiography demonstrated a dilated left coronary artery with a large aneurysm and a small orifice to the right ventricular body. The left ventricular ejection fraction was 55%. Cardiac computed tomography angiography confirmed the diagnosis. Cardiac angiography showed a dilated left coronary artery and a coronary cameral fistula to the right ventricular . The aneurysm was successfully occluded using two Amplatzer™ devices.

Conclusion This case underscores the critical role of multimodal imaging in diagnosing and managing coronary cameral fistulae. Early detection and appropriate intervention are paramount in preventing the progression of symptoms and potential complications such as heart failure and myocardial ischemia. The successful closure with Amplatzer™ devices highlights the efficacy of minimally invasive techniques in treating complex cardiovascular anomalies. Regular follow-up and careful monitoring are essential to ensure long-term success and to manage any potential recurrences.

Clinical key message Timely identification and management of coronary cameral fistulae are crucial to prevent complications. Advances in imaging techniques and minimally invasive treatments, such as transcatheter closure, offer effective solutions. A multidisciplinary approach and regular follow-up are essential for comprehensive care and successful long-term management.

Keywords Coronary artery anomalies, Coronary cameral fistula, Coronary angiography

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Introduction

Coronary cameral fistula (CCF) is a rare congenital anomaly where a coronary artery connects abnormally to a cardiac chamber. Typically arising from embryological abnormalities, CCFs can also result from traumas or surgeries. Congenital coronary anomalies occur in about 1% of the population, with CCFs making up 0.2–0.4% of these cases and appearing in 0.1–0.2% of coronary angiograms [1, 2]. The majority of cases are congenital and solitary, accounting for 0.2–0.4% of all congenital cardiac malformations [3]. These fistulas are often asymptomatic for many years and are frequently diagnosed incidentally during imaging studies [4]. In childhood, asymptomatic cases of CCF are often identified by a continuous heart murmur detected during auscultation. For children older than 3–5 years, elective closure of the fistula is recommended due to the increased risk of complications associated with the condition [5]. In symptomatic cases of CCF, patients may experience manifestations such as angina, exertional dyspnea, syncope, and palpitations. These symptoms arise due to the abnormal blood flow and increased demand on the heart caused by the fistula, which can lead to complications if left untreated [6]. Therapeutic intervention with surgical ligation or transcatheter closure (TCC) is typically performed in patients with symptomatic fistulas or in cases of complications. Percutaneous TCC is preferred because it is less invasive and avoids potential surgical complications. However, surgical closure may be necessary if the vessel is tortuous or if the fistula is large and unsuitable for TCC [7]. TCC of CCF has been advocated as a minimally invasive alternative to surgery [7]. In this study, we report the history of a 5-year-old boy with the diagnosis of coronary cameral fistula, which was treated by an Amplatzer™ duct occluder (ADO).

Case presentation

A 5-year-old Iranian boy presented to the emergency room (ER) with complaints of chest pain and dyspnea on exertion. His medical history revealed multiple similar episodes over the previous 4 months, which had not been evaluated. There was no significant family history of cardiac disease, and his drug history was unremarkable. The patient's social history did not reveal any notable factors contributing to his condition.

On physical examination, his vital signs were stable and within normal ranges. Heart rate was 90 beats per minute, respiratory rate was 20 breaths per minute, blood pressure was 110/70 mmHg, and oxygen saturation was 98% on room air. No murmurs or abnormal heart sounds were detected during auscultation. His cardiopulmonary examination was normal, with clear lung fields and no signs of respiratory distress. Abdominal examination showed no hepatosplenomegaly, and there were no neurological deficits observed.

Methods

The patient was admitted to the pediatric cardiology ward for further evaluation and monitoring. Initial investigations included serial measurements of troponin levels and other relevant blood tests, which are detailed in Table 1. Notably, the lab results revealed mild anemia with a hemoglobin level of 11 g/dL. All other lab results were within normal reference ranges and did not show any remarkable abnormalities. An

Table 1 Laboratory findings of the patient

Test	Result	Reference range
RBC (10^6/μL)	4.6	4.2–5.5
Hemoglobin (g/dL)	11	12–16
WBC (per μL)	9300	4000–11,000
MCV (fL)	89	80–99
Hematocrit (%)	39	37–47
Platelet (per μL)	210,000	150,000–400,000
Neutrophils (%)	64	40–75
Lymphocytes (%)	37.1	20–45
Eosinophils (%)	4.4	0–6
MCH (pg/cell)	32	27–31
MCHC (g/dL)	34	32–36
Troponin I (first sample)	Negative	Negative
Lipid profile, coagulation factors, and troponin		
Cholesterol	144	Up to 200
TG (mg/dL)	91	Up to 150
LDL (mg/dL)	58	Up to 130
HDL (mg/dL)	45	> 45
Blood sugar (mg/dL)	81	74–106
LDH (Iu/L)	340	235–470 Iu/L
K^+ (mEq/L)	4.2	3.5–5.3
Creatinine (mg/dL)	0.4	0.5–1.00
Urea (mg/dL)	11	13–43
HbA1C %Hb	5.2	4.8–5.9
PT (seconds)	11	11–13
PTT (seconds)	30	25–38
INR	1.1	1–1.5
CK-MB	17 u/L	< 25 u/L
Troponin I (second sample in 6 hours)	Negative	Negative

BS blood sugar, *CK-MB* creatine kinase-MB, *Hb* hemoglobin, *HbA1C* glycated hemoglobin, *HDL* high-density lipoprotein, *INR* international normalized ratio, K^+ potassium, *LDH* lactate dehydrogenase, *LDL* low-density lipoprotein, *MCV* mean corpuscular volume, *MCH* mean corpuscular hemoglobin, *MCHC* mean corpuscular hemoglobin concentration, *PT* prothrombin time, *PTT* partial thromboplastin time, *RBC* red blood cell count, *TG* triglycerides, *Urea* blood urea, *WBC* white blood cell count

electrocardiogram (ECG) was performed, revealing normal sinus rhythm with no ST/T wave changes.

Transthoracic echocardiography (TTE) was then conducted, demonstrating a dilated left coronary artery (LCA) with a large aneurysm and a small orifice to the right ventricular (RV) body. The RV size and function were normal, and the left ventricular ejection fraction (LVEF) was measured at 55%. To further delineate the anatomy and confirm the diagnosis, cardiac computed tomography angiography (CTA) was performed, which substantiated the findings of a dilated LCA and coronary cameral fistula to the RV (Figs. 1, 2).

Subsequently, cardiac angiography was conducted, confirming the presence of a dilated LCA and a coronary cameral fistula to the right ventricle. During the procedure, the aneurysm was successfully occluded using two Amplatzer™devices (Fig. 3). Post-procedure angiography of the LCA in the right anterior oblique (RAO) view showed no aneurysmal filling, indicating a successful intervention.

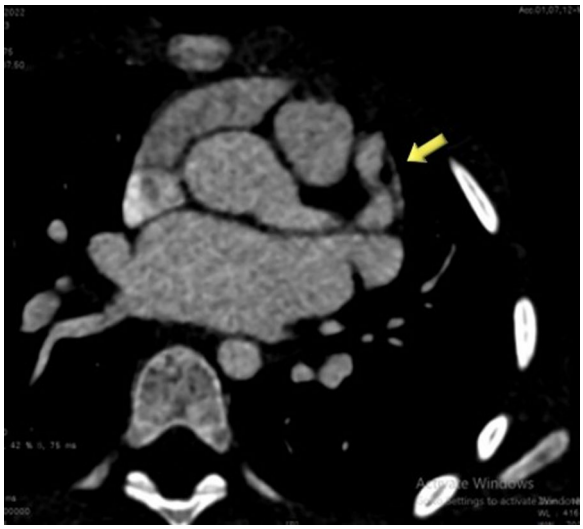

Fig. 2 Cardiac computed tomography angiography (CTA) image showing a coronary cameral fistula (indicated by the arrow). The CTA highlights the connection between the coronary artery and a cardiac chamber, providing a clear view of the anatomical anomaly

Results and follow-up

The day after the angiography, the patient's symptoms were completely resolved. Physical examination revealed normal heart, respiratory system, and neurological function. A chest x-ray was performed, which was normal (Fig. 4). After 48 hours, the patient was discharged without any complications. Follow-up appointments were scheduled at 1, 3, and 6 months after the procedure, during which time the patient reported no abnormal symptoms or recurrences of chest pain or dyspnea.

Discussion

CCFs are rare coronary anomalies characterized by abnormal connections between a coronary artery and one of the cardiac chambers. These and abnormal connections, though uncommon, can have significant clinical implications, including the development of heart failure and non-ischaemic ischemic cardiomyopathy, as evidenced by the patient in this report [1]. In the reported case of a 5-year-old boy, the CCF presented with significant hemodynamic consequences, manifesting as chest pain and dyspnea on exertion, which necessitated

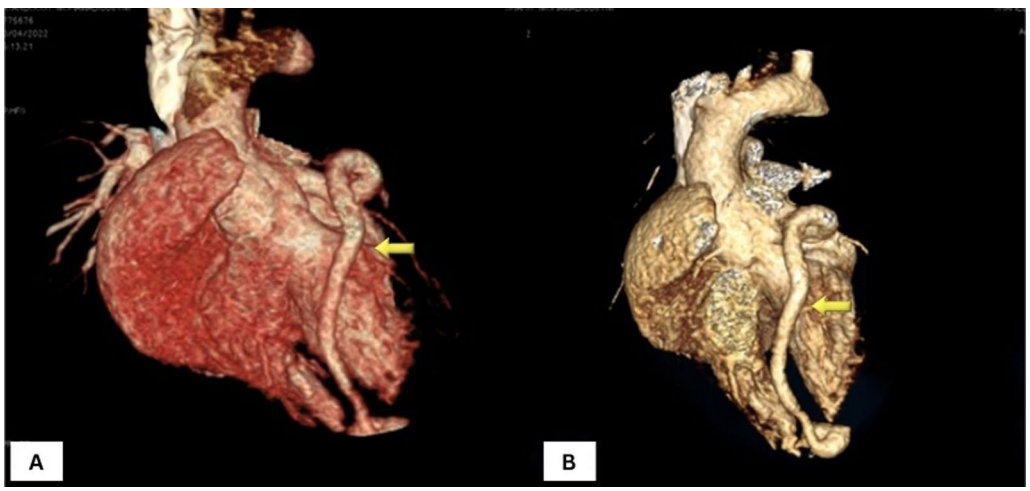

Fig. 1 (**A**) and (**B**) Three-dimensional cardiac computed tomography angiography (CTA) images showing a coronary cameral fistula with a dilated left coronary artery (LCA) and a large aneurysm. Both views (**A** and **B**) illustrate the aneurysmal dilation of the LCA, with the arrows indicating the location of the fistula and aneurysm

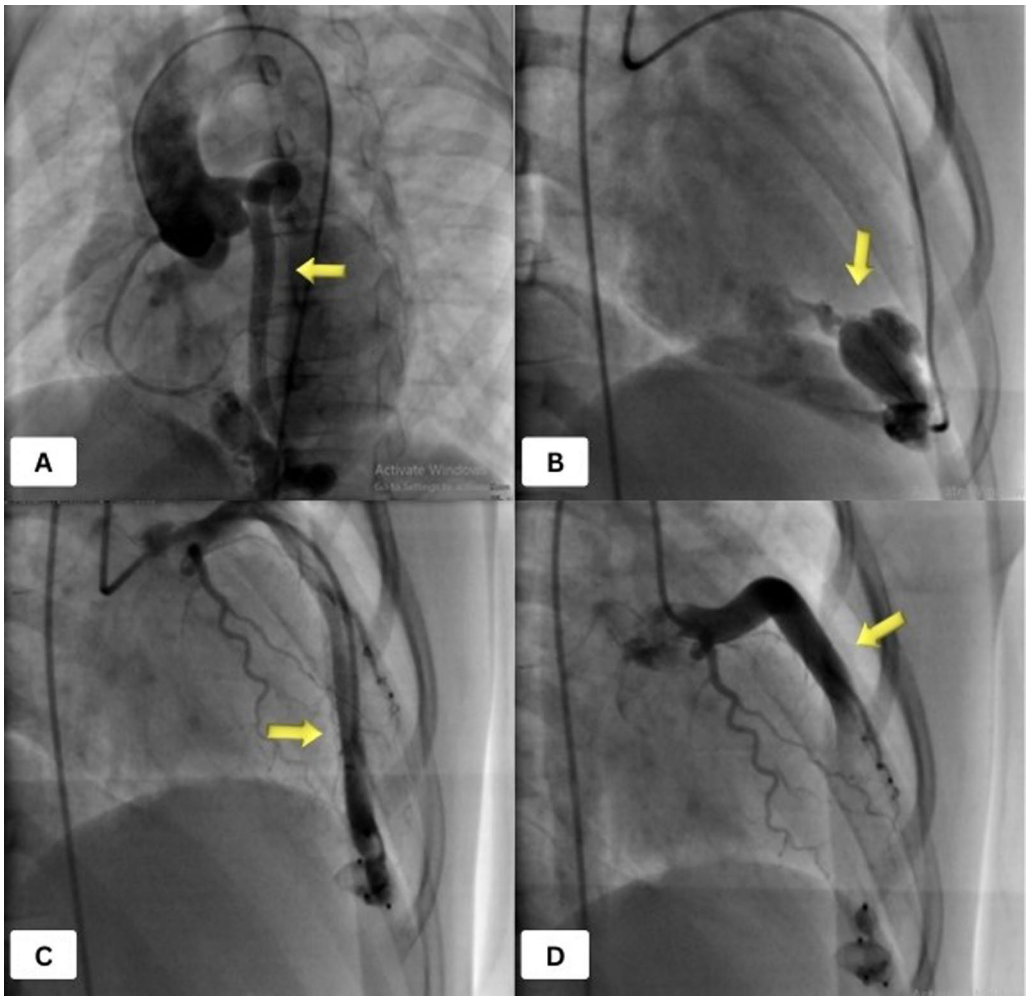

Fig. 3 Cardiac angiography:**A** shows the coronary cameral fistula (indicated by the arrow). **B** illustrates two coronary cameral fistula orifices to the right ventricular (RV) cavity (indicated by the arrow). **C** shows the coronary cameral fistula immediately after the use of the Amplatzer™ device (indicated by the arrow). **D** shows the coronary cameral fistula 2 minutes after using the Amplatzer™ device, with successful occlusion (indicated by the arrow)

prompt intervention with transcatheter closure using Amplatzer™ duct occluders.

Previous studies have shown that the majority of CCF cases are asymptomatic and often discovered incidentally during imaging studies conducted for other reasons. However, symptomatic cases, particularly those with hemodynamically significant shunts, may require intervention to prevent complications such as heart failure and ischemic damage [9] Pathophysiologically, coronary artery fistulas, including CCFs, arise from a failure of the normal development of the coronary vasculature, resulting in a direct connection between a branch of a coronary artery and a cardiac chamber, which can lead to various clinical presentations depending on the severity of the left-to-right shunt [10] These fistulae often lead to a "coronary steal phenomenon," where blood is shunted away from the myocardium, potentially causing myocardial ischemia, particularly during increased myocardial oxygen demand, such as exercise [11].

Diagnostic methods for coronary artery fistulas often begin with noninvasive imaging techniques. TTE is useful for visualizing hemodynamically significant fistulas, while transesophageal echocardiography (TEE) provides detailed information about the origin and insertion point area in adults. Cardiac computed tomography (CT) and magnetic resonance imaging (MRI) offer excellent anatomical delineation, particularly useful for surgical planning [12, 13]. However, coronary angiography remains the gold standard for definitive diagnosis and detailed anatomical assessment of coronary artery fistulas,

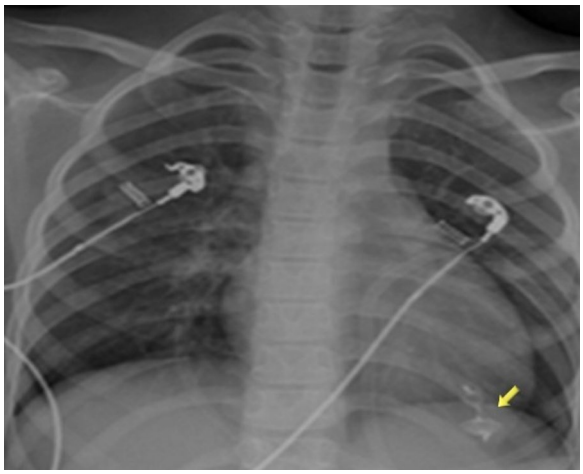

Fig. 4 Chest x-ray taken 1 day after the procedure, showing no signs of complications. The heart and lungs appear normal, and the Amplatzer™ devices are in place and visible (yellow arrow)

providing precise information on the size, course, and drainage site of the fistula [12, 14].

The use of three-dimensional echocardiography has improved the accuracy of anatomical characterization, enhancing the pre-procedural assessment and planning for interventional procedures [15].

Treatment options for CCFs vary on the basis of the clinical significance of the condition and patient comorbidities. According to the updated 2023 American Heart Association (AHA)/American college of cardiology (ACC) guidelines, the management of coronary artery fistulas has evolved to incorporate the latest evidence and a more patient-centered approach. For large fistulas, closure, using either transcatheter embolization or surgical closure, is recommended regardless of symptoms. Smaller fistulas should also be managed with these methods if complications such as ventricular dysfunction, angina, or arrhythmias are present. This approach aims to prevent severe complications and improve patient outcomes by ensuring timely and appropriate intervention [16]. The literature further indicates that transcatheter closure (TCC) is often preferred due to its minimally invasive nature. It avoids complications related to surgical interventions, such as surgical stress, bleeding, infections, and inflammatory responses from cardiopulmonary bypass. In cases where TCC is not feasible due to anatomical challenges, surgical repair remains the most effective treatment. This involves the obliteration of the fistula through epicardial and endocardial ligations. The choice of therapy is influenced by the size, location, and complexity of the fistula, as well as the patient's overall health condition and the presence of comorbid conditions [12, 17]. An innovative stent-assisted coil occlusion technique has also been successfully utilized, providing a promising option for complex cases where traditional methods are not suitable [17].

Complications of untreated CCFs include heart failure, myocardial ischemia, arrhythmias, and endocarditis [11, 18]. The prognosis after treatment is generally favorable, especially with timely intervention. Regular follow-up is essential to monitor for potential recurrence of the fistula or the development of related complications [11]. Continuous follow-up is crucial to monitor for any potential recurrence of the fistula or the emergence of related complications. Moreover, CCFs can lead to increased left ventricular end-diastolic pressure, left ventricular hypertrophy (LVH), congestive heart failure (CHF), atrial fibrillation (AF), ventricular tachyarrhythmias (VT), chronic myocardial ischemia, and myocardial infarction (MI). Hemopericardium can result from the rupture of an associated aneurysm, which is a life-threatening complication [19]. Effective occlusion of CCF can be obtained in more than 90% of cases with the appropriate use of occluder devices, highlighting the importance of careful pre-procedural planning and device selection to minimize the risk of complications [15].

Zenooz *et al.* [4] emphasize the importance of early diagnosis and intervention for CCFs due to the high prevalence of late symptoms and complications, highlighting the role of CT angiography and conventional angiography in the precise delineation of coronary anomalies [10]. Boyle *et al.* [9] also support the use of multimodality imaging for the diagnosis of CCFs, noting that advances in noninvasive imaging have improved the detection and management of these anomalies [9]. Furthermore, Wolking *et al.* [20] highlight the potential for mural thrombosis, rupture, aneurysm formation, and intimal rupture as complications of coronary cameral fistulae, highlighting the importance of careful monitoring and timely intervention [20] Minhas *et al.* [21] also reported that larger fistulas could cause coronary artery steal, resulting in ischemia of the myocardium perfused by the coronary artery distal to the fistula, and other complications such as cardiac failure, atrial fibrillation, bacterial endocarditis, thrombosis, and embolism [21].

CCFs, though rare, are significant anomalies that can lead to severe cardiovascular issues if not treated properly. This case of a 5-year-old boy underscores the importance of prompt diagnosis and intervention. With advancements in imaging technologies such as three-dimensional echocardiography and CT angiography, the accuracy of diagnosing these anomalies and planning for procedures has greatly improved. Transcatheter closure is now the preferred treatment method due to its less invasive nature and high success rate. Continuous follow-up and diligent monitoring are crucial to prevent

Table 2 Literature review of case reports on coronary cameral fistula (CCF)

Author(s) and year	Patient demographics	Presentation and diagnosis	Intervention	Outcome and follow-up
Velázquez-Castañeda *et al*. [22]	7-day-old female	Hx: Dehydration, jaundice, heart murmur; maternal substance abuse. PH/E: Stable, regurgitant murmur at mesocardium. Lab: Not specified. Imaging: TTE and CTA (intercoronary communication between LAD and RCA, coronary ectasia)	Conservative management with medications; f/u scheduled	Asymptomatic; no symptoms reported at 6-month f/u
Moges *et al*. [23]	5-month-old female	Hx: Labored breathing, bluish lips and extremities. PH/E: Tachycardia, central cyanosis, grade I clubbing. Lab: Hypoglycemia, anemia, positive blood culture for coagulase-negative *Staphylococcus*. Imaging: TTE and CTA (large CAF involving left main artery draining into LV, secundum ASD, VSD with right to left shunt, tricuspid valve dysplasia)	Managed in ICU for hypoxic spells; fatal outcome due to respiratory arrest	Infant developed cyanotic spells; succumbed to respiratory arrest despite resuscitative efforts
Srivastava *et al*. [24]	6-year-old male	Hx: Routine cardiac murmur noted in clinic visits. PH/E: NL HR, soft early diastolic murmur at apex. Lab: Not specified. Imaging: TTE, CT, and coronary angiography (dilated LMCA and LAD, multiple fistulous communications to LV, LV noncompaction)	Conservative management with aspirin; regular f/u	Asymptomatic; no intervention required; stress thallium test negative for ischemia
Raj *et al*. [25]	8-year-old female	Hx: Recurrent respiratory tract infection. PH/E: Continuous murmur on auscultation. Lab: Normal blood studies. Imaging: TTE (LCX to RA fistula, dilated LMCA and LCX, dilated RA and RV) and CT angiogram (dilated LMCA with fistulous communication to RA)	Surgical ligation of fistula	Uneventful postoperative course; no residual fistula on f/u echocardiogram at 3 months
Upadhyay *et al*. [19]	20-year-old male	Hx: Fever (8 months), dyspnea on exertion. PH/E: Pulse 106/minute, BP 90/40 mmHg on dopamine, and continuous precordial murmur. Lab: Anemia, positive blood culture for fungal growth. Imaging: TTE (dilated LA, LV; severe aortic regurgitation, vegetations, aortic root abscess) and cardiac CT (dilated sinoatrial nodal artery, pulmonary hypertension, heart failure pattern)	AVR and CCF repair after antifungal treatment	Symptom resolution; successful AVR and CCF repair with no postoperative complications

ASD atrial septal defect, *AVR* aortic valve replacement, *BP* blood pressure, *CAF* coronary artery fistula, *CRP* C-reactive protein, *CTA* computed tomography angiography, *CTCA* coronary computed tomography angiography, *CXR* chest x-ray, *f/u* follow-up, *Hx* history, *LAD* left anterior descending artery, *LCX* left circumflex artery, *LV* left ventricle, *LMCA* left main coronary artery, *NL* normal, *PH/E* physical examination, *RA* right atrium, *RCA* right coronary artery, *RV* right ventricle, *TOE* transesophageal echocardiography, *TTE* transthoracic echocardiography, *VSD* ventricular septal defect, *WBC* white blood cells

and address potential complications such as heart failure, myocardial ischemia, and endocarditis. Effective management of CCFs demands a collaborative approach involving cardiologists, radiologists, and interventional specialists to achieve the best patient outcomes. Similar cases reported in previous literature have been summarized and mentioned in Table 2.

Conclusions

Timely identification of CCFs is essential to prevent severe cardiovascular complications, including heart failure and myocardial ischemia. Advances in imaging techniques, such as three-dimensional echocardiography and CT angiography, have significantly improved diagnostic accuracy and treatment planning. Transcatheter closure has emerged as the preferred treatment modality due to its minimally invasive nature and high success rate, while surgical intervention is reserved for cases where transcatheter closure is not feasible. Regular follow-up and monitoring are crucial to detect and manage potential complications early, such as arrhythmias, myocardial infarction, and endocarditis. A multidisciplinary approach involving cardiologists, radiologists, and interventional specialists is necessary to provide comprehensive care and ensure optimal patient outcomes. Additionally, the use of innovative techniques and devices, such as stent-assisted coil occlusion, offers promising results in treating complex CCF cases where traditional methods pose challenges.

Abbreviations

ADO	Amplatzer™ duct occluder
AF	Atrial fibrillation
ASD	Atrial septal defect
AVR	Aortic valve replacement
BP	Blood pressure
BS	Blood sugar
CAF	Coronary artery fistula
CCF	Coronary cameral fistula
CHF	Congestive heart failure
CK-MB	Creatine kinase-MB
CRP	C-reactive protein
CTA	Computed tomography angiography
CTCA	Coronary computed tomography angiography
CXR	Chest x-ray
ECG	Electrocardiogram
ER	Emergency room
FU	Follow-up
Hb	Hemoglobin
Hb A1C	Glycated hemoglobin
HDL	High-density lipoprotein
HR	Heart rate
ICU	Intensive care unit
INR	International normalized ratio
LAD	Left anterior descending artery
LCA	Left coronary artery
LCX	Left circumflex artery
LDL	Low-density lipoprotein
LDH	Lactate dehydrogenase
LMCA	Left main coronary artery
LV	Left ventricle
LVH	Left ventricular hypertrophy
LVEF	Left ventricular ejection fraction
MCH	Mean corpuscular hemoglobin
MCHC	Mean corpuscular hemoglobin concentration
MCV	Mean corpuscular volume
MI	Myocardial infarction
MRI	Magnetic resonance imaging
NL	Normal
PT	Prothrombin time
PTT	Partial thromboplastin time
RA	Right atrium
RCA	Right coronary artery
RBC	Red blood cell count
RV	Right ventricle
TCC	Transcatheter closure
TEE	Transesophageal echocardiography
TG	Triglycerides
TOE	Transesophageal echocardiography
TTE	Transthoracic echocardiography
VT	Ventricular tachyarrhythmias
VSD	Ventricular septal defect
WBC	White blood cell count

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Author contributions

MT and PE contributed to the conceptualization, methodology, data collection, investigation, curation, writing of the first draft of the manuscript, and submission. HM and MA contributed to the data collection, curation, supervision, software development, and revision of the final draft of the manuscript. ZE and GH contributed to the data collection and the revision of the manuscript.

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Data availability

There is no supporting data, and all available information is included in the manuscript.

Declarations

Ethics approval and consent to participate

The study has been reviewed and approved by the Shaheed Rajaei Cardiovascular, Medical and Research's Ethics Committee. The information provided is anonymized, and the name or characteristics are not mentioned. Therefore, this committee has waived the need for an ethical code. The parent/legal guardian of the patient provided written informed consent on behalf of the patient to participate in this clinical case report. They have been assured that all personal information and medical data will be kept confidential and used solely for research purposes.

Consent for publication

Written informed consent was obtained from the patient's legal guardian for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests

The authors declare no conflicts of interest.

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