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CASE REPORT

Aorta–Right Atrial Tunnel in an Infant

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ABSTRACT: Aorta–right atrial tunnel (ARAT) is an extremely rare congenital heart malformation with an average age of diagnosis of approximately 20 years. Its clinical symptoms are varied and often atypical. ARAT usually originates from the left or right sinus of Valsalva. In this case report, we aim to share a rare case of ARAT originating from the noncoronary sinus in an infant. The patient presented with a cardiac murmur, a dilated right heart, and a tortuous tunnel originating from the dilated noncoronary sinus and terminating at the right atrium in echocardiogram and computed tomography angiography. The patient underwent surgical closure of the tunnel to prevent possible heart failure. Postoperative echocardiography revealed complete closure of the tunnel with no residual flow. No evidence of aortic valve regurgitation or aortic root dilation was detected during 6-month follow-up.

KEYWORDS: ARAT; infant; surgery

1 Case

A 50-day-old boy was referred to our department for a continuous heart murmur. He had a fever and cough 3 weeks prior and was diagnosed with pneumonia when a cardiac murmur was first found. The patient's neonatal pneumonia was successfully resolved following clinical intervention. It is postulated that the onset of this condition may be associated with congenital structural cardiac abnormalities and a persistent left-to-right shunt. The patient was asymptomatic at admission with no sign of growth retardation. Cardiac auscultation revealed a grade 2/6 continuous murmur on the parasternal border. Electrocardiography findings and cardiothoracic ratio were in the normal range.

An echocardiogram revealed a dilated right atrium and ventricle with a normal left ventricular size and function. The aortic valve was tricuspid with mild aortic regurgitation. The noncoronary sinus of Valsalva (NCS) was dilated with a continuous turbulent flow between the NCS and the right atrium, which was approximately 8 mm in diameter. An atrial septal defect (ASD) (10 mm) was detected. To confirm the diagnosis of aorta–right atrial tunnel, a multidetector computed tomography angiography (CTA) was performed. CTA revealed that the left and right coronary arteries and branches were normal. The NCS was enlarged, from which a tortuous tunnel originated and terminated at the right atrial–superior vena cava junction (Figs. 1 and 2).

The patient was diagnosed with aorta–right atrial tunnel (ARAT), which was an extremely rare condition. Considering the obviously dilated right heart detected by both echocardiogram and

CTA, the infant may have already been suffering from abnormal left to right shunt, and was at risk of developing congestive heart failure in near future if the tunnel remained untreated. Therefore, we decided to perform surgical closure of the tunnel. A median sternotomy was performed, and cardiopulmonary bypass with aorto-bicaval cannulation was conducted. The abnormal tunnel was discernible, originating from the aortic root and traveling along the posterior aspect of the right atrium. The tunnel was successfully dissected and looped before cross-clamp without any electrocardiographic change. The aorta was cross-clamped, and cold anterograde cardioplegia was used to arrest the heart. In this way, we were able to open the root of the aorta and directly explore the relationship between the coronary arteries and the aortic opening of the tunnel, as well as the structure of the aortic valve. A transverse incision was made in the aorta to expose the aortic valve and coronary ostium, both of which appeared normal in morphology. Therefore, we decided to close the aortic end of the tunnel directly. Pericardial pledget-reinforced closure was performed at the aortic end to prevent long-term expansion or aneurysm. Right atriotomy was performed, and the atrial end of the tunnel was composed of several small openings, which was consistent with cardiac ultrasound results. Therefore, the tunnel was transected and closed with a polypropylene running suture outside the right atrium. The ASD was closed using a pericardial patch (Fig. 3).

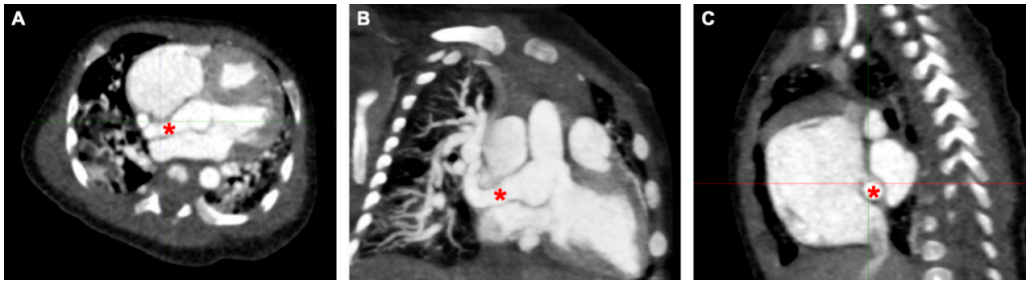

Figure 1: The course of ARAT in computed tomography across different imaging planes. (**A**) Axial view: An anomalous channel arising from the noncoronary sinus of Valsalva, coursing posterior to the right atrium. (**B**) Coronal reconstruction: The channel terminating at the right atrial–superior vena cava junction. (**C**) Sagittal reconstruction. *Aorta–right atrial tunnel.

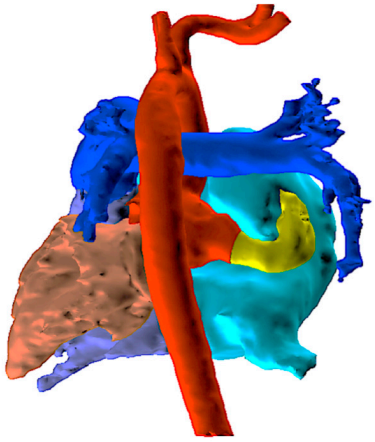

Figure 2: Three-dimensional reconstruction of the cardiac structure based on CT images using the Mimics software, viewed from the posterior aspect. The yellow channel represents ARAT, arising from the dilated noncoronary sinus, coursing posterior to the right atrium. CT, computed tomography; ARAT, aorta–right atrial tunnel.

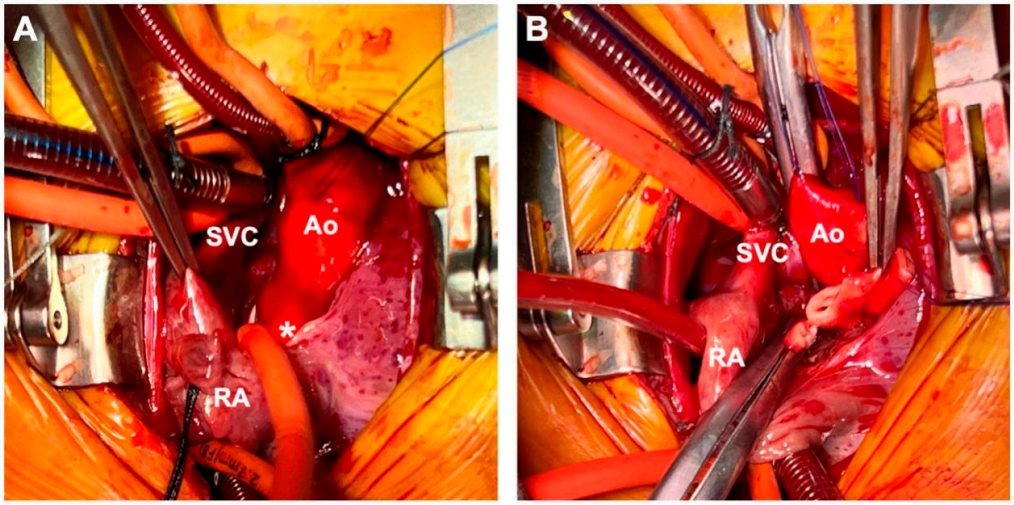

Figure 3: Surgery images. (**A**) ARAT was looped, and the patient's electrocardiogram was monitored for any significant changes before cardioplegic perfusion and aortic cross-clamp. (**B**) The tunnel was transected in the middle part. *ARAT, aorta–right atrial tunnel.

The patient was weaned off ventilation on the second postoperative day and was discharged after 1 week. Postoperative echocardiogram revealed complete closure of the tunnel with no residual flow. During the 6-month postoperative follow-up, both electrocardiographic monitoring and echocardiography showed no evidence of aortic valve regurgitation or aortic root dilation. The infant exhibited age-appropriate growth and developmental milestones, with no documented comorbidities or subsequent hospital admissions.

2 Discussion

ARAT is first described by Coto et al. in 1980 [1]. The embryogenesis of this condition is attributed to a congenital weakness of the elastic lamina in the aortic media [2]. We reviewed all relevant articles and identified a total of 64 cases in 49 publications, most of which were case reports (Table 1). Although the first reported case of ARAT originated from the NCS, this type accounts for the smallest proportion. This case is the fourth reported and the only one in which a cardiac structural abnormality was detected during the neonatal period [1,3,4].

Two-dimensional echocardiography can detect a channel-like structure extending from the aortic root to the right atrium with continuous systolic and diastolic flow on Doppler. Mild to severe enlargement of right heart chambers can manifest. In patients with heart failure, dilated left cardiac chambers and atrioventricular valve regurgitation can be detected. CTA and cardiac catheterization are generally used to confirm the anatomical structure of coronary arteries. Most ARAT cases originate from the left sinus of Valsalva (38/64) and the right sinus of Valsalva (23/64). Clinically, a differential diagnosis should be made with rupture of a congenital sinus of Valsalva aneurysm and coronary cameral fistulas. Rupture of a sinus of Valsalva aneurysm originates mostly from the right sinus of Valsalva and NCS and rarely from the left sinus of Valsalva. Congenital sinus of Valsalva aneurysm has a typical windsock appearance, whereas ARAT usually has an apparent tubular-shaped extra-cardiac course. In our case, the patient was diagnosed during the neonatal period, which suggests that this malformation was a congenital structure instead of a rupture of an aneurysm. Coronary cameral fistula is characterized by the dilation of the

coronary arteries and intra-myocardial coronary artery branches. In patients with ARAT, coronary arteries can be completely normal (46/64), especially when ARAT originates from the NCS. Even if the coronary artery arises from the tunnel, usually adjacent to its origin, the remote part of the ipsilateral coronary artery is always of normal caliber.

The average age of diagnosis was 22.8 ± 18.2 years. A total of 24 patients (37.5%) were asymptomatic at the time of diagnosis. Clinical symptoms ranged from nonspecific ones (such as palpitations, exertional dyspnea, tachypnea, chest pain, and fatigability) to severe ones related to heart failure (such as pedal edema, abdominal distension, and facial puffiness). Although patients can be asymptomatic, surgery is recommended once a diagnosis is made because of the potential long-term risks, including sinus or atrial tachyarrhythmia (eight cases), congestive heart failure (four cases) [5–8], and endocarditis (three cases). The diameter of the channel ranged from 5 to 35 mm, but the degree of shunting depended mainly on the size of the right atrial orifices and if there was a narrowing of the atrial opening. The pulmonary to systemic flow ratio, based on cardiac magnetic resonance imaging (MRI) or cardiac catheterization, varied from 1.1:1 to 6.1:1, with a median of 1.67:1. There were nine patients diagnosed at or under 1 year of age, and four patients underwent surgery during the neonatal period; three of them were prenatally diagnosed, with the earliest diagnosis at 24 weeks of gestation [9–12]. All neonates exhibited severe right atrial and right ventricular enlargement and hemodynamic instability immediately after birth. In this case, the patient had a history of pneumonia during the neonatal period, potentially associated with abnormal shunting, and exhibited significant right heart enlargement upon admission, which warranted early intervention.

Among all patients, 46 patients underwent surgical treatment, 12 underwent percutaneous closure, and 6 were followed up. Surgical methods mainly included direct running sutures, pledgeted closure, and patch closure at the aortic and right atrial ends. The tunnel can also be looped and ligated off-pump. For patients requiring aortic occlusion during surgery, it is crucial to control the channel first to ensure proper and effective perfusion of the myocardial protective solution. For those with coronary arteries originating directly from the channel, multiple techniques were used to reconstruct coronary arteries and the sinus of Valsalva, including reimplantation of the involved coronary artery with or without a patch, closure of the channel distal to the takeoff of the coronary artery, and coronary artery bypass grafting [8,13–18]. Interventional closure devices included vascular plugs, ventricular septal defect and ASD closure devices, duct occluders, and coils. Caution must be exercised to avoid perforation during the procedure, as it may necessitate conversion to mid-sternal surgery [19]. Overall, the surgical outcomes were favorable, with only one early patient death reported [17].

In conclusion, ARAT is an extremely rare congenital heart malformation with an average age of diagnosis of approximately 20 years. Its clinical symptoms are varied and often atypical. ARAT usually originates from the left or right sinus of Valsalva and needs to be differentiated from the rupture of a congenital sinus of Valsalva aneurysm and coronary cameral fistulas. Patients diagnosed during the neonatal period or presenting with clinical manifestations typically exhibit high-volume shunt flow, requiring early surgical intervention. Timely operative management effectively reduces the risk of impending cardiac failure, while potentially preventing long-term complications, such as infective endocarditis and development of atrial fibrillation. The outcomes are generally favorable.

Table 1: General characteristics of patients in different articles.

	Author	Year	Gender	Age	ECG	Associated Lesions	Origin	Coronary Origin	Qp/Qs	Treatment
1	Chigullapalli et al. [13]	2024	F	24 years			RCS	Proximal part of tunnel	2:1	Surgery
2	Dlm et al. [20]	2024	M	14 years			RCS	Proximal part of tunnel		Surgery
3	Ulular et al. [14]	2022	F	27 years	Atrial fibrillation	PFO	RCS	Proximal part of tunnel		Surgery
4	Onorato et al. [21]	2020	M	27 years		Infective endocarditis	RCS	Normal	1.1:1	Occlutech® PDA Occluder
5	Fontes et al. [22]	2019	M	52 years			LCS	Proximal part of tunnel	1.8:1	Follow-up
6	Jain et al. [5]	2018	M	26 years		AI, TR	RCS	Normal		Surgery
7	Narin et al. [9]	2017	M	Neo		PDA	LCS	Normal		Amplatzer Duct Occluder II
8	Dellis et al. [23]	2017	F	38 years		PDA	LCS	Normal		Surgery
9	Khan et al. [15]	2017	M	55 years			RCS	Proximal part of tunnel		Surgery
10	Dingli et al. [24]	2017	F	25 years			LCS	Normal	1.2:1	Follow-up
11	Looi et al. [19]	2016	F	38 years	Atrial fibrillation		LCS	Normal	1.7:1	Surgery
12	Lee et al. [25]	2016	M	31 years			LCS	Normal	1.54:1	Follow-up
13	Kim et al. [26]	2016	M	42 years	Atrial fibrillation	Infective endocarditis, MR, AI	LCS	Normal		Surgery
14	Altiparmak et al. [27]	2015	F	57 years			RCS	Normal	2.2:1	Amplatzer Vascular Plug II
15	Katayama et al. [6]	2015	F	50 years		TR	LCS	Normal		Surgery
16	Venkataraman et al. [7]	2015	F	29 years			RCS	Normal		Surgery
17	Tossios et al. [28]	2014	F	47 years			LCS	Normal	1.5:1	Surgery

Table 1: *Cont.*

	Author	Year	Gender	Age	ECG	Associated Lesions	Origin	Coronary Origin	Qp/Qs	Treatment
18	Iyisoy et al. [29]	2014	F	18 years			LCS	Proximal part of tunnel		Surgery
19	Omeroglu et al. [30]	2013	F	31 years			LCS	Normal	2.5:1	Surgery
20	Kim et al. [31]	2013	F	36 years			RCS	Normal	3.3:1	Surgery
21	Thanopoulos et al. [32]	2013	M	4 years			RCS	Normal		Amplatzer Duct Occluder II
22	Salehi et al. [8]	2013	M	71 years		MR, TR, AI	RCS	Proximal part of tunnel		Surgery
23	Kim et al. [33]	2013	F	42 years			LCS	Normal		Follow-up
24	Baykan et al. [34]	2012	F	3 years		VSD	LCS	Normal	1.88:1	Amplatzer Vascular Plug
25	Winter et al. [35]	2012	F	23 years			LCS	Normal		Amplatzer Septal Occluder
26			M	3 years			LCS	Normal	1.2:1	Follow-up
27	Hu et al. [36]	2012	M	38 years			LCS	Normal		Surgery
28	Matter et al. [37]	2011	F	2 months	Sinus tachycardia		LCS	Normal		Surgery
29	OMaoldomhnaigh et al. [4]	2011	M	8 years			NCS	Normal		Surgery
30	Sung et al. [38]	2011	M	73 years			RCS	Normal	1.4:1	Surgery
31	Myers et al. [39]	2011	M	33 years		Infective endocarditis	LCS	Normal		Surgery
32	Chandra et al. [40]	2011	F	12 years		ASD	RCS	Normal	3:1	Amplatzer Duct Occluder
33	Altekin et al. [41]	2011	M	39 years	Sinus tachycardia		RCS	Normal	1.5:1	Follow-up
34	Krishna et al. [16]	2010	F	11 years			LCS	Normal	1.8:1	Surgery
35			M	24 years			RCS	Normal	1.6:1	Surgery

Table 1: *Cont.*

	Author	Year	Gender	Age	ECG	Associated Lesions	Origin	Coronary Origin	Qp/Qs	Treatment
36	Deshpande et al. [10]	2010		2 days			RCS	Normal		Surgery
37	Subban et al. [42]	2009	F	21 years			LCS	Normal	1.67:1	Amplatzer Duct Occluder
38	Mahesh et al. [11]	2008		4 days			RCS	Proximal part of tunnel		Coils
39	Aggarwal et al. [43]	2007	F	12 years	Sinus tachycardia		LCS	Normal	1.2:1	Surgery
40			M	33 years	Atrial fibrillation		RCS	Proximal part of tunnel	2.7:1	Surgery
41	Al-Hay et al. [44]	2005	M	6 months			LCS	Normal		Surgery
42	Sreedharan et al. [45]	2005	M	11 years			LCS	Normal	1.4:1	Coils
43	Sivakumar et al. [46]	2006	M	19 years			LCS	Proximal part of tunnel	3.2:1	Amplatzer Vascular Plug
44	Gajjar et al. [17]	2005	F	9 years		PDA	LCS	Normal	1.3:1–6.3:1	Surgery
45			F	45 years		ASD	LCS	Proximal part of tunnel		Surgery
46			F	23 years			LCS	Proximal part of tunnel		Surgery
47			M	15 years		ASD	LCS	Normal		Surgery
48			M	18 years			RCS	Proximal part of tunnel		Surgery
49			M	42 years			LCS	Proximal part of tunnel		Surgery
50			M	3 years			RCS	Proximal part of tunnel		Surgery
51			M	10 years			RCS	Normal		Coils
52			F	17 years			LCS	Normal		Surgery

Table 1: *Cont.*

	Author	Year	Gender	Age	ECG	Associated Lesions	Origin	Coronary Origin	Qp/Qs	Treatment
53	Kursaklioglu et al. [47]	2004	M	27 years			LCS	Normal	1.6:1	Surgery
54	Moraes et al. [3]	2004	M	1 year			NCS	Normal		Surgery
55	Türkay et al. [18]	2003	M	29 years			RCS	Proximal part of tunnel	2.1:1	Surgery
56	Tsai et al. [48]	2002	F	2 years			LCS	Normal	1.8:1	Surgery
57	Kalangos et al. [49]	2000	M	18 years			LCS	Normal	1.5:1	Surgery
58			M	7 years			LCS	Normal	1.3:1	Surgery
59	Danilowicz et al. [12]	1989	F	3 days		ASD, PDA	RCS	Normal		Surgery
60	Rosenberg et al. [2]	1986	F	7 years			LCS	Normal		Surgery
61			F	6 months			LCS	Proximal part of tunnel	1.7:1	Surgery
62			M	15 years			LCS	Normal	1.3:1	Surgery
63			M	8 months			LCS	Proximal part of tunnel		Surgery
64	Coto et al. [1]	1980	M	25 years	Sinus tachycardia		NCS	Normal		Surgery

Note: ECG, electrocardiography; RCS, right coronary sinus of Valsalva; LCS, left sinus of Valsalva; NCS, noncoronary sinus of Valsalva; PFO, patent foramen ovale; AI, aortic insufficiency; TR, tricuspid regurgitation; MR, mitral regurgitation; PDA, patent ductus arteriosus; ASD, atrial septal defect; VSD, ventricular septal defect; Qp:Qs, pulmonary to systemic flow ratio.

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Availability of Data and Materials: The data that support the findings of this study are available from the corresponding author, Hao Zhang, upon reasonable request.

Ethics Approval: The study was approved by the Ethics Committee of SCMC (SCMCIRB-K2024198-1), and parental written informed consent was obtained prior to the initiation of the study.

Conflicts of Interest: The authors declare no conflicts of interest to report regarding the present study.

Supplementary Materials: The supplementary material is available online at https://doi.org/10.32604/chd.2025.063108.

Abbreviations

ARAT	Aorta–right atrial tunnel
NCS	Noncoronary sinus of Valsalva

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