

Journal of Clinical and Basic Cardiology

An Independent International Scientific Journal



Journal of Clinical and Basic Cardiology 1999; 2 (2), 279-280

Aborted sudden death during sports activity due to anomalous origin of the left coronary artery from the pulmonary trunk - dilemmas concerning surgical repair of the anomaly

Latific-Jasnic D, Gabrijelcic T, Rakovec P
Ruzic-Medvescek N, Zorman D

Homepage:

www.kup.at/jcbc

**Online Data Base Search
for Authors and Keywords**

Aborted sudden death during sports activity due to anomalous origin of the left coronary artery from the pulmonary trunk – dilemmas concerning surgical repair of the anomaly

D. Latifić-Jasnić, T. Gabrijelčič¹, D. Zorman, N. Ružič-Medvešček, P. Rakovec

A young woman with a previously undiagnosed coronary anomaly – the left main artery arising from the pulmonary trunk – survived ventricular fibrillation which occurred during physical activity (aerobics). A surgical procedure which included the excision of the left coronary artery origin and reimplantation into aorta initially seemed to be successful. Due to the slight twisting of the reimplanted artery, severe stenosis developed and subsequently, bilateral mammary artery anastomosis had to be performed. *J Clin Basic Cardiol* 1999; 2: 279–80.

Key words: coronary anomaly, sudden death, cardiovascular surgery

Anomalous origin of the left main coronary artery from the pulmonary trunk is a rare congenital anomaly. It has a dual natural history [1]: The majority of patients present in infancy with heart failure and echocardiographic signs of dilated cardiomyopathy, 90 % of patients die within the first year of their life; the survivors who reach adult age, however, present with a systemic pulmonary fistula and have a risk of sudden death or congestive heart failure. Sudden death is often exercise-related but is usually not witnessed and seldom heralded by the presence of symptoms or signs [2]. The surgical methods of treatment extend from palliative to radical operations; among the latter, direct reimplantation of left coronary into aorta is preferred if technically possible [3].

Case report

A young woman with described coronary anomaly, who has survived exercise-induced ventricular fibrillation, and in whom two surgical interventions were necessary for complete recovery, forms the basis of this report.

The patient, a 24-year-old woman, underwent the first cardiac catheterization at the age of 5 years because of an systolo-diastolic murmur. Since coronary angiography was not done, the real cause for this murmur was not discovered. Cardiomyopathy, probably due to left ventricular fibroelastosis, was suspected.

The patient's growth and development was since then normal, and other diagnostic procedures were thus not attempted. Because of persisting heart murmur, her cardiac status was reevaluated at the age of 18 years; radionuclide ventriculography revealed mild reduction in her ejection fraction, which remained unchanged during exercise, and anteroapical hypokinesia. In the following years, she was asymptomatic even under rather strenuous physical activities.

In November 1994, she lost consciousness during an aerobics session without any prodromal symptoms or signs. In the first moments shallow breathing was still present, then apnea ensued. Fortunately, a physician attended the same aerobics session and the patient was resuscitated immediately. In approximately 6 to 7 minutes the ambulance had arrived and ventricular fibrillation was confirmed subsequently. She was successfully defibrillated. After a short period of artificial ven-

tilation she began to breathe spontaneously and she regained consciousness. The later clinical course has been uneventful.

Echocardiography showed normal size of the left ventricle with hypokinesia of the anteroapical region. The dilated orifice of the right coronary artery was seen at approximately 12 o'clock in parasternal short axes basal plane. Dilated and tortuous branches of the right coronary artery were seen by 2D imaging and increased flow in them shown by color Doppler flow mapping. A systolo-diastolic flow was revealed by pulsed Doppler. The orifice of the left coronary artery could not be observed clearly; nevertheless, there was no obvious echocardiographic evidence of its anomalous origin, even by transesophageal examination.

Left ventricular angiography showed anterolateral hypokinesia of the left ventricle. No mitral insufficiency was found. Selective coronary angiography confirmed that right coronary artery was very large. Contrast material injected into the right coronary artery filled the left anterior descending and the circumflex arteries via a rich collateral circulation, and a retrograde opacification of the pulmonary trunk became apparent. Blood gas analysis during cardiac catheterization showed an increase in oxygen saturation in the pulmonary artery, and a significant left-to-right shunt.

The patient underwent surgery. The anatomical situation allowed that the ostium of the left coronary artery could be excised from the pulmonary trunk and reimplanted into the ascending aorta. The length of the left coronary artery and its position demanded slight rotation of the reimplanted coronary artery with respect to its longitudinal axis. After the operation, coronary angiography showed a rather normal anatomical situation, except for the fact that the left coronary artery appeared somewhat twisted, but widely patent.

The one-year follow-up coronary angiography, however, revealed substantial narrowing of the twisted segment and demanded reoperation (Figure 1). Two internal thoracic (mammary) artery anastomoses – right thoracic artery on left anterior descending branch and left on first marginal branch of the circumflex system – were performed. Since then, the patient has been well and noninvasive assessment showed normal results.

A repeat invasive study has not been performed because the patient gave birth to a healthy son in August 1997.

Comment

This patient of ours is a very rare case, in whom sudden arrhythmia, evidently cause of sudden death in patients with this anomaly, was witnessed. Exercise can be a trigger for fatal arrhythmia, however, some of these patients tolerate exercise quite well and without angina [4]. Therefore, foreseeing the onset of fatal arrhythmias can be unpredictable.

The evaluation of children with suspected congenital cardiac anomalies used to include cardiac catheterization, but not coronary angiography. In this way, coronary anomalies could not be discovered. We therefore feel that cardiac catheterization in children should be accompanied with coronary angiography in unclear and ambiguous cases. Echocardiography is also of great help by showing the dilated right coronary artery and is in some cases capable of leading to a correct diagnosis without invasive diagnostics [5]; nevertheless, coronary angiography should remain the principal diagnostic tool.

The reimplantation of the left coronary artery into aorta is probably the best surgical option if it can be technically performed without stretching or overtwisting the reimplanted artery [6]. Relatively good initial results can deteriorate with time, therefore careful follow-up is mandatory. In technically less favorable situations, either more sophisticated (eg, tunnel operation) or more conventional operations (eg, internal mammary anastomosis or bypass grafting), may yield better results [7].

References

1. Ortiz De Salzar A, Juanena C, Aramendi JL, Castellanos E, Cabrera A, Agosti J. Anomalous origin of the left coronary artery from the pulmonary artery. *J Cardiovasc Surg* 1990; 31: 801–4.
2. Taylor AJ, Rogan KM, Virmani R. Sudden cardiac death associated with isolated congenital coronary artery anomalies. *JACC* 1992; 20: 640–7.
3. Tkebuchava T, Carrel T, von Segesser L, Real F, Jenni R, Turina M. Repair of anomalous origin of the left coronary artery from the pulmonary artery without early and late mortality in 9 patients. *J Cardiovasc Surg* 1992; 33: 479–85.

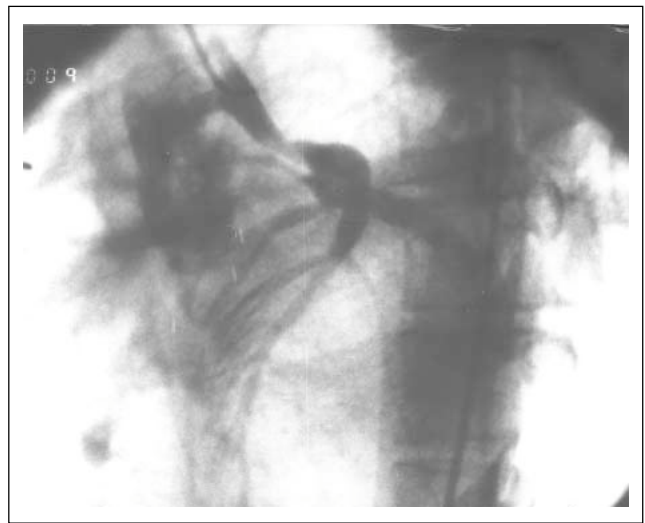


Figure 1. Angiogram of the left coronary artery after the first operation. The left main coronary artery is twisted and substantially narrowed.

4. Viskin S, Belhassen B, Paredes A, Yakirevich V, Miller H, Laniado S. Anomalous origin of the left coronary artery from the pulmonary artery: report of an adult with ventricular fibrillation as the presenting symptom. *Am Heart J* 1992; 124: 221–2.
5. Koike K, Musewe N, Smallhorn JF, Freedom RM. Distinguishing between anomalous origin of the left coronary artery from the pulmonary trunk and dilated cardiomyopathy: role of echocardiographic measurement of the right coronary artery diameter. *Br Heart J* 1989; 61: 192–7.
6. Alexi-Meskishvili V, Hetzer R, Weng Y, Lange PE, Jin Z, Berger R, Loebe M. Anomalous origin of the left artery from the pulmonary artery. Early results with direct aortic reimplantation. *J Thorac Cardiovasc Surg*. 1994; 108: 354–62.
7. Backer CL, Stout MJ, Zales VR, Muster AJ, Weigel TJ, Idriss FS, Mavroudis C. Anomalous origin of the left coronary artery. A twenty-year review of surgical management. *J Thorac Cardiovasc Surg* 1992; 103: 1049–58.

Mitteilungen aus der Redaktion

Besuchen Sie unsere zeitschriftenübergreifende Datenbank

☒ [Bilddatenbank](#)

☒ [Artikeldatenbank](#)

☒ [Fallberichte](#)

e-Journal-Abo

Beziehen Sie die elektronischen Ausgaben dieser Zeitschrift hier.

Die Lieferung umfasst 4–5 Ausgaben pro Jahr zzgl. allfälliger Sonderhefte.

Unsere e-Journale stehen als PDF-Datei zur Verfügung und sind auf den meisten der marktüblichen e-Book-Readern, Tablets sowie auf iPad funktionsfähig.

☒ [Bestellung e-Journal-Abo](#)

Haftungsausschluss

Die in unseren Webseiten publizierten Informationen richten sich **ausschließlich an geprüfte und autorisierte medizinische Berufsgruppen** und entbinden nicht von der ärztlichen Sorgfaltspflicht sowie von einer ausführlichen Patientenaufklärung über therapeutische Optionen und deren Wirkungen bzw. Nebenwirkungen. Die entsprechenden Angaben werden von den Autoren mit der größten Sorgfalt recherchiert und zusammengestellt. Die angegebenen Dosierungen sind im Einzelfall anhand der Fachinformationen zu überprüfen. Weder die Autoren, noch die tragenden Gesellschaften noch der Verlag übernehmen irgendwelche Haftungsansprüche.

Bitte beachten Sie auch diese Seiten:

[Impressum](#)

[Disclaimers & Copyright](#)

[Datenschutzerklärung](#)