



Contents lists available at ScienceDirect

American Journal of Emergency Medicine

journal homepage: www.elsevier.com/locate/ajemThe
American Journal of
Emergency Medicine

Pediatric out-of-hospital cardiac arrest caused by left coronary-artery agenesis with primary shockable rhythm. A brief report

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ARTICLE INFO

Article history:

Received 8 March 2017

Received in revised form 9 May 2017

Accepted 10 May 2017

Available online xxxx

Keywords:

Pediatric

Cardio-pulmonary resuscitation

Out of hospital cardiac arrest - shockable rhythm

Automated external defibrillator

Coronary artery agenesis

ABSTRACT

Background: To illustrate a rare cause of out-of-hospital cardiac arrest in children, its differential diagnoses, emergency and subsequent treatment at various steps in the rescue chain, and potential outcomes.

Case presentation: A 4-year-old boy with unknown agenesis of the left coronary ostium sustained out-of-hospital cardiac arrest. Bystander cardio-pulmonary resuscitation was initiated and defibrillation was performed via an automated external defibrillator (AED) shortly after paramedics arrived at the scene, restoring sinus rhythm and spontaneous circulation. After admission to the intensive care unit the child was intubated for airway and seizure control. Further diagnostic work-up by angiography revealed agenesis of the left coronary artery. After initial seizures, the boy's neurological recovery was complete. He subsequently underwent successful internal mammary artery in-situ bypass surgery to the trunk of the left coronary artery. One year after cardiac arrest, the patient had completely recovered with no physical or intellectual sequelae. A catheter examination proved excellent growth of the bypass and good cardiac function.

Conclusions: This case illustrates the long term outcome after agenesis of the LCA while reiterating that prompt access to pediatric defibrillation may be lifesaving—albeit in a minority of pediatric OHCA.

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1. Background

Non-traumatic, unspecific out-of-hospital cardiac arrest (OHCA) in children is a rare event, dreaded by paramedics and emergency physicians alike [1]. Reported incidence rates vary from 3 to 20 per 100,000 person-years. [8–11, 16–19].

Typically, collapse occurs just minutes after symptom onset [2] and even the most skilled teams may have little experience with prehospital

Abbreviations: AED, Automated External Defibrillator; AHA, American Heart Association; ALCAPA, Anomalous left main coronary artery from the pulmonary artery; CPR, Cardio-Pulmonary Resuscitation; ECG, Electrocardiography; LCA, Left Coronary Artery; LMCA, Atresia of the Left Main Coronary Artery; OHCA, Out-of-Hospital Cardiac Arrest; OPALS, Ontario Pre-Hospital Advanced Life Support; SIDS, Sudden Infant Death Syndrome; VF, Ventricular Fibrillation; VT, Ventricular Tachycardia.

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resuscitation in this setting. According to estimates in Germany, emergency physicians encounter only one case of pediatric prehospital resuscitation in nine years [3]. Efforts were made to reach performance benchmarks of the American Heart Association (AHA) with regard to chest compression frequency and depth [4].

In cardiovascular-related OHCA, congenital abnormalities are the predominant cause in age groups 0 to 2 years (84%) and 3 to 13 years (21%) [5]. Coronary artery anomalies are the second most common cause of OHCA in the United States, and the second most common cause of sudden cardiac death in young athletes [2,6]. The prevalence of coronary artery anomalies is about 1% in the general population [6], and 2.2% in children with certified cardiovascular death [7]. Approximately half of all cases occur in patients without known heart disease [7].

Coronary artery anomalies are classified as to whether they cause obligatory, exceptional, or no ischemia. The most common congenital coronary artery anomaly is an aberrant outflow of the left main coronary artery from the pulmonary artery (ALCAPA). Because of low vascular resistance in the pulmonary artery, blood flows retrogradely from the right coronary artery via collaterals [8].

<http://dx.doi.org/10.1016/j.ajem.2017.05.011>

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Please cite this article as: Weigeldt M, et al, Pediatric out-of-hospital cardiac arrest caused by left coronary-artery agenesis with primary shockable rhythm. A brief report, American Journal of Emergency Medicine (2017), <http://dx.doi.org/10.1016/j.ajem.2017.05.011>

An anomalous origin of the left main coronary artery from the right sinus of Valsalva is commonly associated with sudden cardiac death [1, 8,9], caused by an obstruction of the coronary artery between the aorta and the pulmonary trunk in combination with a kinking or intrinsic narrowing at the anomalous ostium. Patients with this coronary variant tend to be asymptomatic until sudden cardiac death [8].

Atresia of the left main coronary artery (LMCA) is a rare condition but almost always associated with ischemia [6]. Again, blood flows retrogradely from the right coronary artery, and from smaller towards larger vessels [10,11,12]. This increases the risk of myocardial ischemia

and possible cardiac arrest in situations with enhanced metabolic need. Without symptoms like fatigue, angina, or exercise-induced syncope, however, patients are unlikely to undergo imaging and become diagnosed before any critical or life-threatening event [2].

2. Case presentation

This is a report on an apparently healthy 4-year old boy who, after 2 h of intensive play and exercise at an indoor playground, suddenly complained of stomach pain. Minutes after symptom onset he became

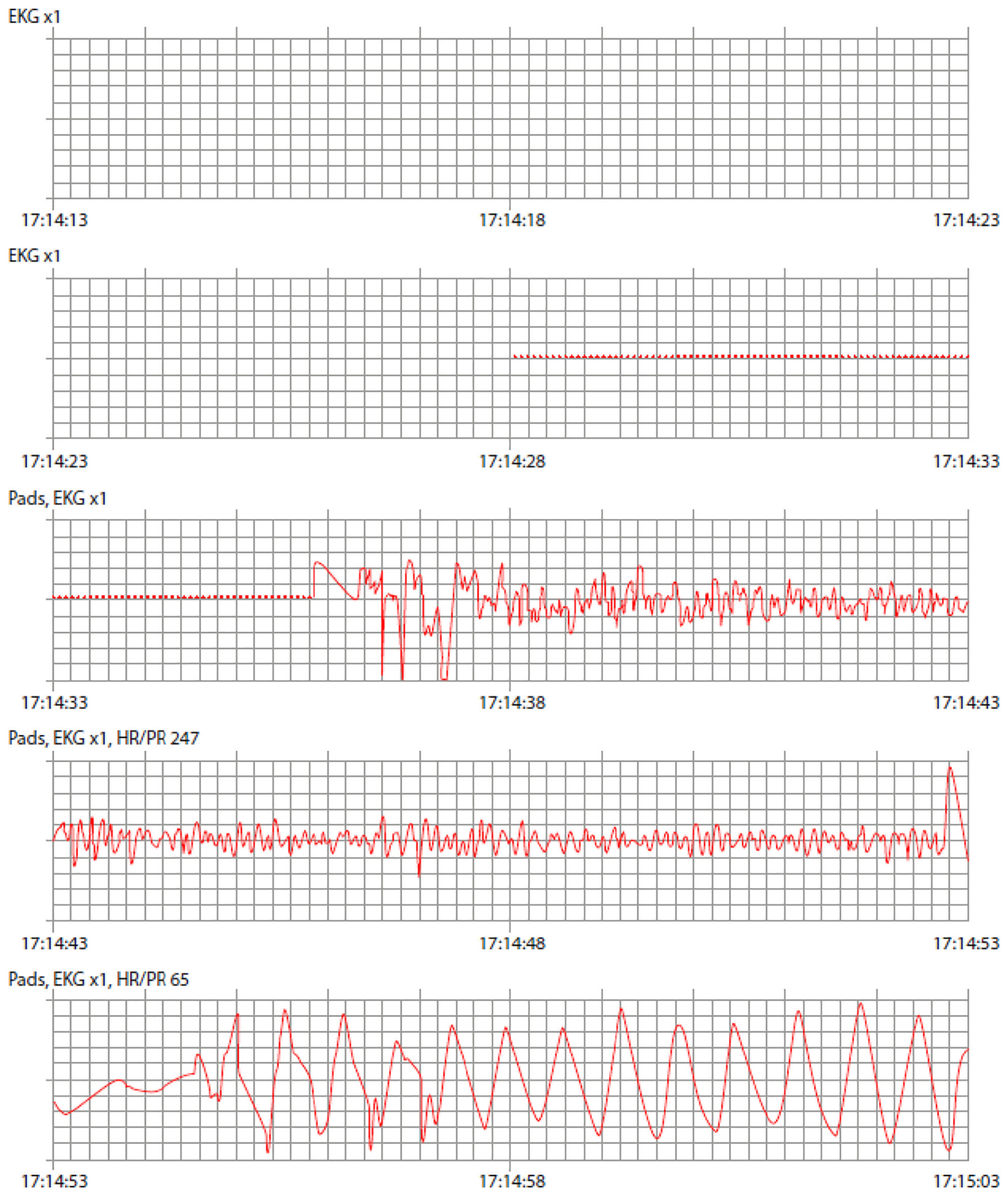


Fig. 1. ECG from AED with applied shock at 17:14:53 and subsequent restored sinus rhythm.

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