Case Report

Lazarus phenomenon in a patient with Duchenne muscular dystrophy and dilated cardiomyopathy

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Abstract

Lazarus phenomenon (LP) is the spontaneous return of circulation after cessation of resuscitation. We herein report the case of a 21-year-old man with Duchenne muscular dystrophy and dilated cardiomyopathy who was hospitalized in the intensive care unit because of respiratory distress. Three days after the initial admission, the patient experienced asystole and did not respond to resuscitation for 30 minutes. Ten minutes after cessation of resuscitation, normal sinus rhythm with a palpable pulse was noted, and the patient was reintubated. Sixty days after admission, the patient was discharged from hospital with complete neurological recovery. Intensive care physicians should be aware of the LP, although it is rare, and a watchful waiting period is advisable after cessation of resuscitation.

Keywords: dilated cardiomyopathy; Duchenne muscular dystrophy; Lazarus phenomenon; spontaneous return of circulation

1. Introduction

Lazarus phenomenon (LP) is the term applied to spontaneous return of circulation after the cessation of cardiopulmonary resuscitation (CPR). The term was first coined by Bray in 1983 because of its resemblance to the Biblical story of Lazarus. The frequency of the condition is not known, and it is considered quite rare because of the scarcity of reported cases. The exact mechanism underlying LP is not known, although hyperventilation, hyperkalemia, late effects of inotropic drugs, and myocardial ischemia are possible explanations. Because of the presumed infrequency of this condition, there are no studies that have specifically researched LP in the scientific literature.

In this article, we report a 21-year-old man with Duchenne muscular dystrophy and dilated cardiomyopathy who experienced in-hospital cardiac arrest and did not respond to cardiac resuscitation for 30 minutes. Ten minutes after cessation of resuscitation, spontaneous respiration and circulation were observed, with a complete neurological recovery by discharge. To the best of our knowledge, this is the first reported case of LP in a patient with dilated cardiomyopathy.

2. Case report

A 21-year-old man with known Duchenne muscular dystrophy was admitted to emergency department with shortness of breath and peripheral cyanosis for 2 days. His only known medical condition was Duchenne dystrophy and he was not using any medications. On physical examination, his blood pressure was 100/70 mmHg, pulse rate was 120 beats/minute, and body temperature was 35.9°C. Auscultation revealed...
globally reduced pulmonary sounds, with a 1/6 systolic murmur at the apex of heart. His electrocardiogram (ECG) showed sinus rhythm with mild tachycardia and prominent R waves in the right precordial leads (Fig. 1). He was cachectic, with severe muscle wasting in all extremities (Fig. 2). Arterial blood gas analysis showed severe, uncompensated respiratory acidosis with the following measurements: pH, 6.9; partial pressure of O₂ (pO₂), 74 mmHg; partial pressure of CO₂ (pCO₂), 105 mmHg; and HCO₃, 22.5 mmol/L. He was diagnosed with respiratory failure and possible pneumonia, transferred to the intensive care unit (ICU), and intubated to avoid a respiratory collapse. His initial biochemical analysis showed hypokalemia (2.68 mEq/L) with high liver transaminase levels (alanine aminotransferase, 292 IU/L; aspartate aminotransferase, 109 IU/L). His initial medical treatment included the following medications: cefazolin [3 × 1 g, intravenously (i.v.)], salbutamol [6 × 2.5 mg intratracheal (i.t.)], N-acetylcysteine (10 mL; 10% sol.; 6 × 1 i.t.), famotidine (50 mg 3 × 1 i.v.). Potassium chloride was given through a central line to correct hypokalemia, but the rate of infusion was kept slow to avoid cardiac arrest. The initial echocardiographic assessment showed mild left ventricular dilatation, moderate systolic dysfunction with an ejection fraction of 35%, and right ventricular dilatation with normal arterial pressure (Fig. 3). Three days after hospitalization, short bursts of polymorphic tachycardia were observed on telemetry, which rapidly deteriorated to sinus bradycardia and asystole. He was resuscitated for 30 minutes. During resuscitation, ventilation was maintained with the mechanical ventilator in the intermittent mandatory ventilation mode. A total of 11 mg of adrenaline was given during resuscitation. Resuscitative efforts were halted after 30 minutes as no electrical activity was seen on the ECG. Mechanical ventilation was stopped and the endotracheal tube was removed. Five minutes after the cessation of CPR, an ECG showed asystole without any electrical activity (Fig. 1). However, the monitor electrodes were not removed from the patient, and 10 minutes after cessation of resuscitation a nurse noticed sinus rhythm on the monitor. As his pulse was palpable, he was intubated and mechanical ventilation was reinstituted. His blood pressure was 109/90 mmHg, and an ECG obtained a few hours after resuscitation showed findings similar to the initial ones (Fig. 1). Blood gas analysis obtained after stabilization of the patient showed mild acidosis with pH, 7.35; pCO₂, 40 mmHg; pO₂, 125 mmHg; and HCO₃, 22 mmol/L. Biochemical analyses obtained within 24 hours of recovery revealed persistently elevated transaminase levels and normal potassium levels (K, 4.01 mEq/L), while his renal function tests and urinary output were normal (creatinine, 0.14 mg/dL; blood urea nitrogen, 15 mg/dL; calculated creatinine clearance, 295.14 mL/minute). The patient regained consciousness 6 hours after resuscitation, and he was capable of understanding questions within 8 hours. A repeat echocardiogram showed findings similar to the initial one, with an ejection fraction of 25–30% and mild mitral regurgitation (Fig. 3). At follow up, his spontaneous breathing was inadequate for extubation, and a tracheostomy was performed on the 33rd hospital day (Fig. 2). During his hospitalization in the ICU, pseudomonas pneumonia and candidemia were diagnosed, and treated. On the 60th day of hospitalization, the patient was discharged with a home-type ventilator.

3. Discussion

As far as we know, this case is the first demonstration of LP in a patient with an underlying dilated cardiomyopathy. After asystolic cardiac arrest, patients with advanced heart disease are less responsive to CPR and have poorer outcomes compared
with those without underlying cardiac conditions. Despite these setbacks, this case shows that a delayed response to CPR (or a spontaneous recovery) is still possible in these patients. In addition, we observed complete recovery of higher cortical functions after 10 minutes of cardiac arrest. Although interesting, this latter finding has been reported before with other LP cases, making it an attractive target for research.

LP was first described in the medical literature in 1982, but it has possibly been known for decades. Since then, at least 40 cases of LP have been reported in the medical literature, although it is suggested that LP remains underreported because of medicolegal concerns. Some cases have been reported only in newspapers and in similar nonscientific publications. As the number of reported cases are few and represent a heterogeneous population (ranging from acute myocardial infarction to recreational drug use), no unifying theories exist to explain LP. Suggested explanations are inflation of the lungs due to hyperventilation, late effects of drugs (when given through a peripheral route), hyperkalemia, and the effects of ischemia. However, there was at least one reported LP patient who did not have any of these conditions. Even in our patient, these factors were not present, as ventilation was maintained mechanically during resuscitation, the drugs were administered through a central line, and the patient had hypokalemia rather than potassium excess.

A unique feature of this case was the presence of dilated cardiomyopathy, which was associated with muscular dystrophy. LP has been reported after an acute cardiac event (myocardial ischemia/infarction), which was the most frequently identifiable cause of LP in a comprehensive review. This latter finding may reflect the frequency of cardiac arrests due to myocardial ischemia, rather than a pathophysiologic link. Spontaneous or interventional relief of ischemia in acute myocardial infarction is associated with dramatic improvement in prognosis. Because ischemia does not play a prominent role in asystole associated with nonischemic dilated cardiomyopathy, this case presents a unique “cause” of LP. Patients with dilated cardiomyopathy and heart failure are more prone to asystole/pulseless electrical activity, and have a less favorable outcome after CPR, so it would be logical to assume that spontaneous return of circulation after cessation of resuscitation would be highly unlikely in this patient population.

Nevertheless, this case shows that such recoveries can occur in these patients against all odds. Therefore, a watchful waiting period of 10–15 minutes after cessation of CPR is advisable in all patients with cardiopulmonary arrest, even when the underlying cardiac condition is not reversible.

Partial or complete neurological recovery after LP has been reported previously. In their comprehensive review, Adhiyaman et al reported that 45% of patients who experienced LP had partial or complete neurological recovery. Of note, their review data indicated that nine patients had cessation of circulation (without any efforts at resuscitation) for more than 5 minutes. Six of the nine patients (66%) had partial or complete neurological recovery.

Fig. 2. Photographs of the patient after cardiac arrest. (A) Picture taken 2 days after cardiac arrest. The picture was taken while the patient was asleep. Note the atrophy in the extremity muscles. (B) Photograph 1 month after resuscitation. The patient was able to understand verbal commands perfectly at that time.

Fig. 3. Echocardiogram of the patient taken before cardiac arrest. Still images of the left ventricle during (A) diastole and (B) systole. Left ventricular wall motion is reduced, accompanied by mild dilatation. Note that the right ventricle is also dilated. The still frames were obtained from a modified parasternal long axis view because of postural abnormalities in the patient.
complete neurological recovery. We also observed a similar phenomenon in our patient, with a rapid and complete recovery of higher cortical functions. There is a striking resemblance between these cases and hibernation, where some types of mammals can survive in conditions close to cardiac arrest without neurological damage. However, until there is more evidence from experimental studies, the latter idea will remain as a highly speculative hypothesis considering that the available neurological recovery data are from a small-scale review of a few case reports.

Practically, watchful waiting of 10–15 minutes after cessation of CPR, preferably with continuous monitoring of cardiac electrical activity, could be beneficial for a minority of patients, even for those with cardiac failure. The unexpected neurological recovery of this patient (and previously reported cases) after a considerable duration of complete cardiac arrest stresses the need for studies on this phenomenon, as understanding of LP could lead to advances that could be beneficial for all cardiac arrest patients.

Conflicts of interest

The authors declare no conflicts of interest related to this work.

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