Postoperative Breath-Holding Spells Requiring Cardiopulmonary Resuscitation After Cardiopulmonary Bypass

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Abstract
Breath-holding spells are common in childhood and can be associated with bradycardia and pulselessness. This report details severe breath-holding spells complicating postoperative management after atrial septal defect closure. The patient required cardiopulmonary resuscitation despite the use of a temporary pacemaker to prevent bradycardia. After multiple episodes of chest compressions, the decision was made to not intervene immediately to pulselessness and the patient was able to recover without further intervention.

Keywords
cardiopulmonary resuscitation, breath-holding spells, cardiopulmonary bypass, pediatrics

Introduction
Breath-holding spells (BHSs) are common in childhood and occur in roughly 4% to 5% of the general pediatric population, typically from the ages birth to 4 years.¹,² Breath-holding spells involve a sustained pause in the expiratory phase of respiration and possibly loss of consciousness. Spells are classified as pallid, cyanotic, or mixed.³ Precipitating factors for cyanotic BHSs include anger and frustration, whereas pallid BHSs often follow surprise, such as an unexpected injury.¹ Cyanotic BHSs are indicative of cortical overdrive of respiratory muscles, as demonstrated by breath-holding to the threshold of cerebral hypoxia. Pallid BHSs have a component of vagal mediation and, as a result, are associated with bradycardia and asystole. The exact mechanism by which this autonomic dysregulation occurs is unknown.³ Episodes are self-resolving, no long-term repercussions are evident, and most patients outgrow the spells by 5 years of age.²

While BHSs are largely addressed in an outpatient general pediatric setting by providing parental reassurance, severe BHSs after cardiac surgery may initially result in an aggressive response. This case report will detail pulseless pallid BHSs in a young child after atrial septal defect (ASD) closure. Four of these spells resulted in cardiopulmonary resuscitation (CPR). The father of the patient consented to the publication of this article.

Case Report
A 22-month-old male was admitted to the pediatric Cardiac Intensive Care Unit (CICU) following an uncomplicated修补 repair of a secundum ASD. The patient’s past medical history was notable for failure to thrive and developmental delay with possible undiagnosed genetic disorder. Parents later provided a history of spells where he would hold his breath and become pale, but did not lose consciousness. Cardiopulmonary bypass time was 23 minutes and the postoperative transesophageal echocardiogram showed good biventricular function with no residual defects. A clear prime was utilized; the patient received no blood products. He was extubated in the operating room and returned to the CICU on low flow supplemental oxygen at 2 LPM. Sedation and analgesia were achieved with a dexmedetomidine infusion at 0.5 µg/kg/h, scheduled intravenous acetaminophen, and as needed intravenous morphine.

The first BHS occurred 80 minutes after CICU admission and was provoked by chest tube stripping. The patient was noted to become agitated, paused in expiration, and turned gray. He did not improve with stimulation, blow-by oxygen, or bag-valve-mask (BVM) ventilation. Despite these interventions, the patient’s saturations dropped to the 30s, his heart rate decreased to 48 beats per minute, and no pulse was felt (Figure 1). At this time, chest compressions were initiated. The patient was found

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to have a pulse with an adequate blood pressure after 30 seconds of compressions; no rescue medications were given. A pacemaker was attached to the temporary epicardial pacemaker wires and set to VVI mode at a rate of 100 beats per minute.

Within an hour, the patient once again held his breath and peripheral oxygen saturations decreased below 40%; BVM ventilation was started. He developed bradycardia and appropriate ventricular pacing was seen. Despite pacing, the patient was found to have absent palpable pulses that correlated with a flattened arterial waveform (Figure 2). The patient received compressions for 20 seconds, after which time he had palpable pulses with an appropriate arterial line waveform. The dexmedetomidine infusion was stopped and a propofol infusion was ordered at 20 µg/kg/min. During the setup of the propofol infusion, he had another BHS with desaturation and hypotension. The BVM was utilized along with a bolus of propofol and the patient recovered.

The third CPR event occurred 2 hours later; it was thought to be provoked by the patient’s agitation with his nasal cannula. The BVM ventilation was started; however, it was not felt to be effective as the patient was fighting the ventilatory assistance. The patient subsequently became bradycardic, causing the pacemaker to start pacing. Again, the patient’s arterial line dampened with an imperceptible pulse. He received 60 seconds of CPR, after which he was intubated. Fentanyl and dexmedetomidine infusions, at rates of 2 and 0.3 µg/kg/h, respectively, were started after intubation. An echocardiogram was obtained to evaluate the surgical repair; the images showed the repair was intact with normal biventricular function.

The patient remained sedated and intubated without incident for the following 12 hours. The next morning, the patient woke and became agitated. No end-tidal carbon dioxide reading was registered, the arterial waveform had dampened, and the ventilator was alarming for high peak inspiratory pressures with minimal return tidal volumes. Desaturation with this event was mild with a saturation recorded of 75%; however, the arterial line flattened and CPR was started. Epinephrine 0.005 mg/kg was administered, which resulted in spontaneous return of circulation after 60 seconds of CPR. The patient was paralyzed with rocuronium and sedated with 1 mg of Ativan. An epinephrine infusion was started for hypotension thought to be related to increased sedation medication use. The dexmedetomidine infusion was discontinued and a Versed infusion was started at 1 µg/kg/min. Another echocardiogram was performed with no concerning findings.

Four hours later, the decision was made to attempt extubation with one-on-one care from a cardiac intensivist. A deep extubation was performed after giving 3 mg of propofol to avoid a BHS associated with extubation. As sedation infusions were decreased, hypotension resolved and the epinephrine infusion was discontinued. After waking, the patient continued to have intermittent BHSs for several hours, with desaturations and loss of arterial waveform, although CPR was deliberately not done. All spells resolved within 2 minutes and decreased in frequency throughout the day. The patient remained in the CICU for an additional day for observation and no further spells occurred during the remainder of the hospitalization. At outpatient follow-up 5 weeks after
surgery parents reported that he was still having mild BHS but they were “not as impressive” as before surgery.

Discussion

The child in this case report had a classic presentation of pallid BHSs, including desaturation, bradycardia, and loss of consciousness. Uniquely, these spells occurred during the postoperative period with invasive hemodynamic monitoring and close observation by a highly trained CICU nurse. This context and timing confounded the approach to the best intervention and, perhaps, complicated care. While the above events were quickly identified as BHSs, the team could not guarantee that BHSs were solely responsible for the hemodynamic changes observed when postoperative aberrations were possible. A multidisciplinary review of the case was conducted and all were in agreement with the interventions enacted, including the utilization of CPR.

While BHSs with severe bradycardia and asystole have been detailed in the literature, reports remain scarce about spells in the postoperative period. Resolution of pallid BHSs, including those with asystole, occurs spontaneously without CPR, although these spells are distressing for caregivers.2,4 Given the likelihood of recovery without intervention, the team decided not to perform further CPR, even with the loss of pulses following extubation. It was assumed that further interventions would only aggravate the patient and lead to continued BHSs.

There are reports of pacemaker implantation decreasing the frequency of BHSs in cases when bradycardic or asystolic events affect quality of life.5-8 Interestingly, even with a temporary pacemaker in place, the patient still developed pulselessness. The same autonomic dysfunction that resulted in bradycardia may have also decreased the patient’s systemic vascular resistance, which was poorly tolerated after cardiopulmonary bypass. Also, with one of the spells, he only desaturated to 75%, yet still became pulseless. This suggests that hypoxia was not the only mechanism for his hemodynamic instability.

Appropriate BHS evaluation remains undefined. An electrocardiogram (ECG) should be obtained, especially for pallid BHSs, for the purpose of excluding long QT syndrome as an alternative diagnosis. This patient did have an ECG pre- and postoperatively, which did not indicate long QT syndrome.

Interestingly and perhaps coincidentally, this patient received a blood transfusion of postoperative day 2 for a hematocrit of 23.9% due to acute blood loss related to the perioperative period. After this transfusion, he had no subsequent severe BHSs. The patient did not have a history of anemia and had a normal hemoglobin and hematocrit preoperatively (11.4 g/dL and 33.3%). Iron deficiency has been found concomitantly in children who have BHSs. Randomized control trials have shown improvement upon initiation of iron supplementation in both children with anemia and without.2,9,10 The blood transfusion could be responsible for his decrease in spell severity after discharge.

Summary

We present the case of a child whose post-bypass care was complicated by pulseless BHS. Invasive hemodynamic monitoring and protocols for resuscitation led to multiple episodes of CPR. The patient fully recovered and was able to discharge from the ICU when a strategy of nonintervention was employed.

Author Contributions

All authors contributed to the design of this case report and were involved in final editing. All authors approve of the final manuscript.

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Ethics Approval

Institutional review board (IRB) approval was waived for this case report.

Informed Consent

Family of the patient agreed to publication of this case report.

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