Intraoperative Cardiac Arrest Due to the Oculocardiac Reflex and Subsequent Death in a Child with Occult Epstein-Barr Virus Myocarditis

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FATAL cardiac arrest due to the oculocardiac reflex (OCR) is uncommon. Myocarditis is also infrequent, although well known, cause of sudden death. This entity may go unrecognized in patients whose illness may resolve spontaneously, thus the true prevalence of myocarditis in the general population is unknown. In several major textbooks, myocarditis is not mentioned as a cause of either morbidity or mortality related to anesthesia. We describe the case of a boy who died after OCR-induced cardiac arrest during strabismus surgery and who was found to have Epstein-Barr virus (EBV) myocarditis.

Case Report

A 5-yr-old, 18.8-kg boy was taken to surgery for elective correction of right-sided strabismus. He had undergone two similar surgical procedures uneventfully when he was 17 and 48 months old. His medical history was otherwise unremarkable, with no known allergies. Two weeks before surgery, he had symptoms of an upper respiratory tract infection but was asymptomatic and afebrile afterward. There was no family history of sudden death, cardiac dysrhythmia, or other cardiac disease.

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pancuronium (0.1 mg/kg) were administered intravenously, and moderate hyperventilation (carbon dioxide tension 30 mmHg) with 100% O₂ was instituted.

Two and a half hours after the end of surgery, the patient was transferred to the pediatric intensive care unit for monitoring. On arrival, he was hemodynamically stable. His Glasgow coma score was 6. On physical examination, a low-grade (1/VI) systolic murmur was audible over the right and left sternal borders. The liver was enlarged, with the edge palpable at 3 cm below the right costal margin; the tip of the spleen was felt at 1-2 cm below the left costal margin. The trachea remained intubated in the postoperative period because of persistent coma. No other significant hemodynamic event was noted in the intensive care unit. Heterophile antibodies were present using a rapid slide agglutination test. The Paul-Bunnel-Davidsohn assay was diagnostic for EBV-induced heterophile antibodies: agglutination at a titer of 1:512 in unabsorbed serum and at a titer of 1:1792 and 1:28 after absorption with guinea pig kidney and beef red cells, respectively. Specific immunoglobulin M antibodies to EBV-capsid antigen were positive by ELISA. Further laboratory studies were as follows: ASAT 82 IU/L, ALAT 30 IU/L, CPK 314 IU/L, cytokomavirus IgG and IgM antibody negative by ELISA, and normal cerebrospinal fluid (erythrocytes nil, leukocytes nil, protein 0.13 g/L, glucose 4.73 mmol/L). Blood, urine, and cerebrospinal fluid cultures were negative. Cardiac echo-Doppler studies were normal. Electrocadioagrams remained normal throughout the patient's stay in the intensive care unit. Auditory and visual evoked potentials and cerebral computed tomography results were normal. However, 24 h after surgery, an electroencephalogram showed pronounced slow diffuse activity. The Glasgow coma score remained at 1-2 for 5 days and then deteriorated progressively until brain death was declared on the 5th postoperative day.

Postmortem examination revealed chronic inflammation in all three cardiac layers. The inflammatory infiltrate was composed mostly of lymphocytes and activated lymphocytes with a few plasma cells and rare eosinophils; superimposed on this diffuse picture were tiny foci of severe inflammation surrounding degenerated segments of myocardial fibers. Moderate inflammation was observed in the liver, spleen, and mediastinal and abdominal lymph nodes. Severe anoxic brain edema was found with hernia of the temporal unci and cebellar amygdala.

Discussion

In the current case, postmortem histologic findings were compatible with a systemic viral infection. Current or recent EBV infection was demonstrated by the presence of a heterophile antibody profile specific for infectious mononucleosis,12-14 and confirmed by the presence of EBV-specific Ig M. No evidence of infection of the central nervous system was found pre- or post-mortem. These observations indicate that our patient had an active EBV infection, with death due to an acute anoxic encephalopathy secondary to an intraoperative cardiac arrest. We think that this arrest was related to two major factors acting simultaneously: an OCR exacerbated by an EBV myocardiitis.

The initial traction on the lateral rectus muscle at the beginning of surgery immediately induced bradycardia followed by a ventricular rhythm and circulatory arrest. The frequency of the OCR during correction of strabismus ranges from 30% to 90%, depending on the definition used for a positive response to stimulation.15,16,17 The incidence of transient, nonfatal cardiac arrest lasting a few seconds may be as high as 2.4 per 1,000 ophthalmic operations.18 However, in almost all cases, stopping the surgical stimulus is sufficient to re-store spontaneous sinus rhythm, and only one previous specific case of death directly attributable to the OCR has been reported.17 Thus, despite the high incidence of the OCR during strabismus surgery, mortality related to this type of surgery is low. Cooper et al. reported a mortality rate of 1.1 per 10,000 strabismus corrections in the 0-19-year-old age group, with 11 deaths occurring in almost 100,000 operations performed.18 In our own institution, approximately 20,000 corrections of strabismus were done in children during the past 20 yr, and the patient described in this case report was the only fatality. Furthermore, this child had undergone two previous strabismus corrections without incident. We believe that a second factor, i.e., the underlying subclinical myocardiitis, was responsible for the circulatory arrest. It has been suggested that subjects with cardiac disorders are prone to develop complications due to the OCR19; our report supports this assumption.

Myocarditis as a cause of death associated with anesthesia has been reported only once, in a 35-year-old woman.20 This is surprising for many reasons. Although acute myocarditis usually is considered an uncommon cause of sudden unexpected death,2 myocarditis may occur but remain unrecognized in many patients whose illnesses resolve spontaneously21; the prevalence of "silent" myocarditis may be greater than generally suspected.22 Acute myocarditis may be the primary cause of death resulting from either a fatal arrhythmia or a cardiogenic shock.23 Furthermore, one would expect myocarditis to significantly increase the risk of cardiac arrest and mortality related to general anesthesia, especially because of its dysrhythmic potential. The fact that, to date, only two anesthesia-related deaths have been attributed to myocarditis—this woman22 and our patient—may be due, in part, to the controversy over the histologic criteria for the diagnosis of myocarditis.24


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However, the most likely explanation may be that almost all cases of myocarditis severe enough to cause sudden death during anesthesia are not clinically silent but are sufficiently apparent to lead to postponement of elective surgery.

The current case, with the observed association of an exaggerated OCR leading to cardiac arrest and death in a child with an occult EBV myocarditis, although exceptional, illustrates that asymptomatic myocarditis may preexist in patients presenting for general anesthesia. There is no satisfactory screening test for occult myocarditis. It has been recommended that wider use be made of routine preoperative ECG tracings but there has been no large-scale prospective study on the utility of such a practice. Normal ECG tracings may be observed even in the presence of severe myocardial damage in the acute phase of viral myocarditis.22 Furthermore, ECG findings such as ST segment or T-wave changes and PR segment or QT interval prolongation do not necessarily reflect myocardial injury.21 In conclusion, clinical screening does not suffice to eliminate completely the risk of having a patient with “silent” viral myocarditis under general anesthesia. The risk of mortality related to the OCR should be considered low but not absent, despite sophisticated monitoring and even in experienced hands.

References


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