

reviewed by anesthesiologists to alert them to potential child abuse by parents.

This case is the only report of delayed awakening from anesthesia due to probable child abuse, although child abuse masquerading as other illness has been reported.⁶⁻⁸ Other causes of delayed awakening include the anesthetic agent,⁹ hypoxia, hypercarbia, hypoglycemia, electrolyte derangements, hypothermia, other drugs, and surgical complications. Of the numerous factors that may account for delayed awakening, the anesthesiologist should be aware that preoperative or postoperative undocumented or unauthorized drug administration are possible etiologic factors.

In summary, the goal of the "family unit" concept of care is to involve the family as much as possible in the care of the hospitalized child. Our case demonstrates that the family cannot universally be relied on to provide care and accurate information. Because of this observation, we recommended: 1) screening the parents for child abuse before utilizing the "family unit" concept of pediatric hospital care; 2) monitoring parents when administering any substance to their hospitalized child; and 3) obtaining a drug screen on patients who exhibit unexplained or unusual patterns of awakening from anesthesia.

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Malignant Hyperthermia in Duchenne Muscular Dystrophy

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Duchenne muscular dystrophy (DMD) is the most severe and rapidly progressive form of the common

muscular dystrophies.¹ Muscle biopsy is an essential part of the laboratory evaluation and usually must be obtained during general anesthesia because the patients are usually pediatric patients.

Anesthesia-related cardiac arrest² and rhabdomyolysis^{3,4} have been described in patients with DMD. Rowland⁵ recently suggested that these cases^{2,3} may also have malignant hyperthermia (MH), although this interpretation has been questioned by Rosenberg.⁶ Willner *et al.*⁷ also suggested that MH may occur in patients

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with DMD but provided no evidence to support their claim. In this report, we describe a case of probable MH in a child with DMD. The diagnosis of MH subsequently was confirmed by use of the caffeine contracture test.

REPORT OF A CASE

A 5-year-old boy was admitted for dental extraction under general anesthesia. No unusual features were noted on examination.

After premedication with 20 mg promethazine, po, anesthesia was induced with halothane and 70% nitrous oxide. Succinylcholine, 20 mg (1 mg/kg), was then given iv. Masseter spasm followed and the patient's arms failed to relax. Intubation of the trachea was accomplished with difficulty because of this spasm. Shortly thereafter, widening of the QRS complex on the ECG was noted, which was followed by an idioventricular rhythm and finally ventricular fibrillation. Cardiac resuscitation was initiated and maintained for approximately 40 min; normal cardiac function then returned; Defibrillation was successful on the sixth occasion. During resuscitation, 120 mEq sodium bicarbonate, 20 mg lidocaine, 200 mg calcium gluconate, and 0.5 mg adrenaline were given iv. Because the episode was considered clinically to be possible MH, 20 mg dantrolene was given iv as soon as it was available (the cardiac rhythm had just been restored), and a second dose was repeated 45 minutes later.

All anesthetic gases were discontinued and ventilation was controlled with a Fi_{O_2} of 1.0. The analysis of arterial blood gases, potassium, and calcium data are listed in the table 1. The profound metabolic acidosis noted 5 and 10 minutes post-arrest was considered to result from the hypermetabolic state, and the respiratory acidosis was considered secondary to an inadequate compensatory increase in ventilation. Creatine kinase (CK) drawn one hour post-resuscitation was greater than 64,000 units (normal 0–225). Reddish-colored urine, which reacted positively with orthotolidine and did not contain red blood cells (presumptive evidence of myoglobinuria), was noted for 24 hours. At no time during the anesthetic or resuscitation did the patient show hyperpyrexia. However, in spite of being completely exposed in a cool operating room and having received cold iv solutions, his rectal temperature did not fall below 36° C.

Two days post-arrest, the child was weak but walked with assistance. He was discharged home 8 days post-arrest and was clinically well. A specimen of serum drawn before the anesthetic showed a CK level of 24,000 units. These results were not available preoperatively.

Subsequent inquiry disclosed that, although the patient's developmental motor milestones were normal, his parents had noted that he appeared awkward, fell easily, did not run well, and had trouble getting up off the floor. The family history was negative for MH or muscular dystrophy.

Physical examination two months later revealed a slight lumbar lordosis, mild calf muscle hypertrophy, mild quadriceps muscle atrophy, mild pelvic and shoulder girdle weakness, a positive Gower's sign (rising from a stooped position by climbing up the legs using the hands), myopathic electromyogram (short duration, polyphasic, low amplitude, easily recruited muscle action potentials), and a CK level greater than 4,800 units. These clinical and laboratory features are typical of DMD.¹

At this time, a muscle biopsy specimen was removed from the vastus lateralis muscle for *in vitro* contracture⁸ and histopathologic⁹ studies performed according to previously described techniques, under general anesthesia. On this occasion the patient was premedicated with diazepam po and meperidine im. Fentanyl, droperidol, and diazepam were given iv for induction of anesthesia. Anesthesia was maintained with nitrous oxide and fentanyl. Ventilation was controlled. Temperature and cardiac rate were monitored continuously. The general anesthetic and post-anesthetic courses were uneventful.

TABLE 1. Table of Laboratory Values

	Minutes Post-arrest		Minutes Post-resuscitation	
	5	10	10	60
Arterial blood gases				
pH	6.94	6.94	7.10	7.46
P _{O₂} (mmHg)	19.00	12.00	60.00	369.00
P _{CO₂} (mmHg)	74.00	80.00	27.00	31.00
O ₂ Saturation (%)	—	—	—	97.00
Bicarbonate (mm/l)	15.00	17.00	0.50	22.00
Base excess (mm/l)	−16.00	−14.00	−22.00	0.00
Calcium (8.5–10.5 mg/dl)	5.60			
Potassium (3.5–5 mEq/l)		8.70		6.60

In Vitro Caffeine Contracture Test and Histopathology

The caffeine specific concentration (CSC), defined by Britt *et al.*¹⁰ as the concentration of caffeine required to induce a one-gram contracture in a strip of muscle, was calculated for all specimens. The base line tension for contracture studies was one gram. Control CSCs for 29 patients in our laboratory have ranged from 3.5–10 mM. In duplicate muscle specimens from this patient, CSC values of 2.95 and 2.70 mM caffeine were obtained, thus indicating that the patient had malignant hyperthermia.

When halothane was added to the test situation, CSCs of 0.15 and 0.19 mM were obtained. In controls (number 29) we have never obtained CSCs less than 0.3 mM. Although the results in this patient are abnormal when halothane was added, in our laboratory, CSCs obtained in the presence of halothane have not been as consistently a reliable indicator of MH susceptibility as CSCs obtained with caffeine alone.

The muscle had the following abnormalities: variation in fiber diameters, internalization of nuclei, fiber splitting, large opaque fibers, clusters of degenerating and regenerating fibers, and increase in endomysial fibrous and fatty connective tissue. This pathologic picture is classic for DMD.¹¹

DISCUSSION

Contracture studies of individuals who have survived a clinical episode of MH have shown that an abnormal contracture response is diagnostic of clinical MH.^{8,12} Similarly, an abnormal contracture response is considered the most reliable indicator of MH susceptibility when evaluating persons who may be at risk for MH.¹³ In our patient, we concluded that the abnormal contracture response supported the opinion that the clinical episode, while lacking some of the features of MH, was in fact MH rather than some other problem such as ventricular fibrillation secondary to hyperkalemia.

Perhaps in patients with DMD, contracture tests cannot be used to confirm an episode of MH or to determine MH susceptibility. This question will not be answered until there are sufficient reports of contracture responses in DMD patients. Moulds *et al.*¹⁴ reported normal contracture responses in two patients with myotonia congenita and three patients with myotonic dystrophy. We previously found normal contracture responses in two patients with myotonic dystrophy. These

observations suggest that the presence of muscular dystrophy does not in itself make the contracture response abnormal, and that an abnormal contracture response can be used to indicate MH susceptibility even in patients with muscular dystrophy.

There is a widespread impression, supported by Richard's report¹⁵ of 61 uncomplicated general anesthetics in 43 patients with DMD, that general anesthesia in DMD, is normally uneventful. Our case report proves that MH may complicate general anesthesia in this group of patients. While this is the first report of MH in DMD proven by contracture studies, there are two other published reports of probable MH in patients with DMD,² but neither patient had the diagnosis confirmed by contracture studies. We believe that MH must now be recognized as a possible complication of general anesthesia in DMD patients.

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Hypertension after Intraoperative Autotransfusion in Bilateral Adrenalectomy for Pheochromocytoma

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Management of patients with pheochromocytoma includes preoperative alpha-adrenergic receptor blockade, intraoperative blood pressure control, and intravascular volume expansion with crystalloid solutions and

blood.¹ We describe a patient with bilateral pheochromocytomas who was also a Jehovah's Witness,² but who consented to intraoperative use of autotransfusion. Concomitant with the transfusion of autologous packed erythrocytes, a dramatic increase in systemic arterial pressure was observed.

REPORT OF A CASE

A 31-year-old woman with the syndrome of multiple endocrine neoplasias, type II, including bilateral pheochromocytoma was admitted for bilateral adrenalectomy. She had previously undergone bilateral radical neck dissection for thyroid medullary carcinoma. The patient was a Jehovah's Witness and would not accept transfusion of any banked blood products. However, she did consent to the intraoperative use of autotransfusion using a Cell-Saver® (Haemonetics Cell

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