

# Rhabdomyolysis as a Postoperative Complication of Multilevel Soft-tissue Surgery in a Child With Cerebral Palsy

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Rhabdomyolysis is a potentially life-threatening syndrome if unrecognized. The most common causes are trauma, excessive muscle activity, alcohol abuse, and toxic substances. Rhabdomyolysis as a postoperative complication in children with cerebral palsy who have received multilevel soft-tissue surgery has not been reported in the literature. The purposes of this study are to present the case of a 12-year-old boy with spastic quadriplegic cerebral palsy who developed rhabdomyolysis after soft-tissue release and to review the literature. The patient was treated with adequate sedation and hydration, and discharged in a stable condition 11 days after surgery. His serum creatine kinase level had returned to within the normal range by the 17<sup>th</sup> postoperative day. At the 6-month follow-up, there were no systemic sequelae. The prompt recognition of rhabdomyolysis depends on a high level of suspicion. Routine checks of urine color after surgery is mandatory. For patients with high muscle tone, monitoring of muscle enzymes is recommended. Adequate sedation, pain control and hydration may prevent the progression of this life-threatening condition. [*J Chin Med Assoc* 2010;73(12): 651–654]

**Key Words:** cerebral palsy, postoperative complications, rhabdomyolysis

## Introduction

Rhabdomyolysis is a syndrome resulting from skeletal muscle injury. The severity varies widely, from asymptomatic elevation of muscle enzymes to life-threatening complications of acute renal failure (ARF) or electrolyte imbalance.<sup>1</sup> Prompt recognition and early intervention are crucial to a better outcome. The causes of rhabdomyolysis include crush injury, overexertion, alcohol abuse and certain medicines, inherited genetic disorders, metabolic disorder and infections.<sup>2</sup> Rhabdomyolysis in children with cerebral palsy (CP) after soft-tissue surgery has not been reported in the literature. The purposes of this report are to present a case of rhabdomyolysis after multilevel soft-tissue surgery in a child with quadriplegic CP, and to review the literature.

## Case Report

A 12-year-old boy with spastic quadriplegic CP (Gross Motor Function Classification System: level V) presented to the orthopedics clinic due to poor sitting balance and deformity of multiple joints. Delayed motor milestones had been noted since infancy, and CP with spastic quadriplegia was diagnosed. This child also had language and communication dysfunction. The patient's weight was 21 kg, height was 121 cm, and body mass index was 14.34 kg/m<sup>2</sup>. Physical examination revealed positive Thomas test in both hips, with flexion contracture of 20°. He had hip adduction contracture with an abduction angle of 30° in each hip. The popliteal angle measured 80° in both knees. The Silfverskiold test revealed tight gastrocnemius muscle bilaterally.



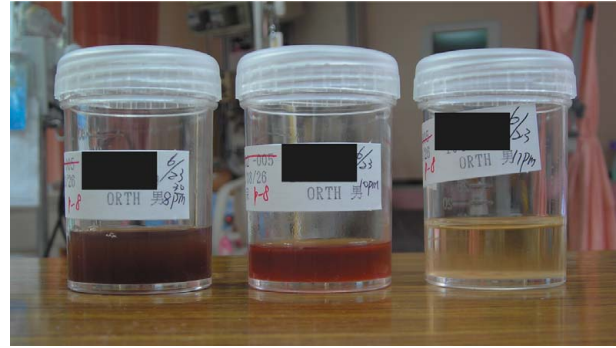
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Single-event multilevel surgery including bilateral over-the-brim iliopsoas lengthening, adductor longus and gracilis muscle release, hamstring lengthening, and gastrocnemius recession was performed. The patient was placed in the supine position and adequately padded. The patient received anesthetic medications including intravenous atropine, atracurium, thiamylal, neostigmine and inhaled sevoflurane. The whole operative and anesthetic course went smoothly. Total operative time was 320 minutes. There was no abnormal temperature elevation, no unstable vital signs, no abnormal muscle contracture or other complications.

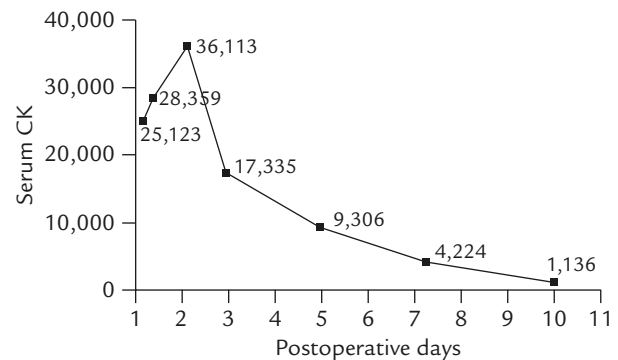
On the 1<sup>st</sup> day after surgery, from the morning onwards, the boy was agitated and hypertonic. Analgesics, including morphine 2 mg every 4 hours, were given, but persistent hypertonic status and agitation were still noted. At the 25<sup>th</sup> postoperative hour, he was found to have tea-colored urine and decreased urine output. On physical examination, the boy showed no focal cranial nerve abnormality or excessive swelling of the limbs, but there was marked increase of muscle tone. Laboratory tests showed serum creatine kinase (CK) of 25,123 U/L, and the muscle and brain component (MB) of CK was 403 U/L (ratio, 1.6%). Serum chemistry revealed sodium of 148 mmol/L, potassium of 4.8 mmol/L, blood urea nitrogen of 10 mmol/L (28 mg/dL), and creatinine of 88.4  $\mu$ mol/L (1.0 mg/dL). Hemogram showed a white blood cell count of  $15.9 \times 10^9$ /L (15,920/mm<sup>3</sup>), hemoglobin of 6.45 mmol/L (10.4 g/dL), and platelet count of  $267 \times 10^9$ /L (267,000/mm<sup>3</sup>). Prothrombin time, international normalized ratio, and partial thromboplastin time were all within normal limits. Urinalysis revealed dipstick positive for hemoglobin, with microscopic examination positive for 15–18 red blood cells per high-powered field. Under the impression of rhabdomyolysis, the boy was admitted to the intensive care unit (ICU) for further management.

During the ICU stay, adequate sedation, pain control, intravenous fluid supplement and intermittent diuretics were given. His urine color returned to normal during his 1<sup>st</sup> day in the ICU (Figure 1). His CK level peaked in the 51<sup>st</sup> hour after surgery and then decreased gradually (Figure 2). His urine output was maintained within the 3–5 mL/kg/hr range. Sleeping electroencephalography (EEG) revealed no abnormal findings.

The patient was discharged in a stable condition 11 days after surgery. His serum CK level had returned to within the normal range by the 17<sup>th</sup> postoperative day. At the 6-month follow-up, the patient's contractures remained corrected, and there were no systemic sequelae from rhabdomyolysis.



**Figure 1.** Tea-colored urine was observed on the 1<sup>st</sup> postoperative day, which gradually returned to normal color after medical treatment.



**Figure 2.** Serum creatine kinase (CK) level peaked on the 2<sup>nd</sup> postoperative day and declined gradually in the following days.

## Discussion

Rhabdomyolysis is the final common pathway of many different conditions. The skeletal muscles are injured and release muscle contents into the plasma. Myalgia, weakness and red-to-brown-colored urine are the classic symptoms. The definitive diagnosis of rhabdomyolysis requires an elevation of CK levels to more than 5 times of normal in the absence of significant elevation of brain or cardiac CK fractions.<sup>3</sup> The syndrome can result in severe complications. Severe hyperkalemia and hypocalcemia have a potential effect on cardiac arrest, and muscle swelling can cause compartment syndrome.<sup>4</sup> However, the most dangerous sequela of rhabdomyolysis is ARF, which is associated with multiorgan failure in children with rhabdomyolysis.<sup>5</sup> The exact mechanism of ARF is unclear but may be attributable to vasoconstriction, hypoperfusion, renal tubular dysfunction, cast formation, and myoglobin-induced tubular toxicity.<sup>2–5</sup> The associated rate of ARF in adults has been estimated to be between 17% and 35%.<sup>3</sup>

Laboratory examination is necessary to confirm the diagnosis. Serum levels of myoglobin rise quickly and return to normal 1–6 hours after muscle injury ends,

due to the rapid renal clearance of myoglobin. Thus, CK level has become the preferred laboratory marker to detect and monitor rhabdomyolysis. Serum CK level begins to rise 2–12 hours after the insult and peaks 24–72 hours later.<sup>4</sup> In rhabdomyolysis, the urine dipstick test is strongly positive for blood, which indicates pigmenturia. But only a few red blood cells can generally be identified in the 100× microscopic field.

Surgery can cause CK levels to elevate. Yousef et al<sup>6</sup> reported a range of CK elevation after major surgeries of 4–647 U/L. The surgery itself is less likely to elevate the CK value as high as 25,123 U/L, which was the level reached in our case.

Rhabdomyolysis is a documented complication of many surgeries, such as neurosurgeries, bariatric surgeries, orthopedic surgeries (with use of tourniquets), and urological surgeries. Muscle compression, long-term immobilization and tissue ischemia were the most common causes; long operative time and morbid obesity are major risk factors.<sup>7–10</sup> General anesthesia has also been reported to be associated with rhabdomyolysis when complicated with malignant hyperthermia or propofol infusion syndrome.<sup>11,12</sup> Since our patient was not obese, limb position had been changed frequently during the operation, a tourniquet was not used, and the anesthesia course was smooth, the above-mentioned surgery-related factors were not found in this case.

The reported nonsurgical causes of acute pediatric rhabdomyolysis include cold weather, heat stroke, infection, intoxication, venom, trauma, metabolic derangement, status asthmaticus, hereditary conditions, excessive muscle activity as seen in convulsions, extreme exertion, dystonia and spastic posturing.<sup>4</sup> Our patient had no known environmental exposure, intoxication, infection or trauma. The negative history of prior rhabdomyolysis and no recurrent episode during follow-up exclude metabolic or hereditary causes in our patient.

Excessive muscle activity might be the cause of rhabdomyolysis in this case. According to the caregiver's description, the patient was used to contracting his muscles in response to physical discomfort, which is inevitable after surgery (postoperative pain, immobilization in cast, etc).

Clinically, it is difficult to differentiate hypertonia from seizure, especially in a child with communication dysfunction. The most reliable test to detect epilepsy is simultaneous EEG, which is not clinically practical. A normal post-ictal EEG cannot exclude seizure. An epilepsy attack is less likely to cause a large amount of muscle damage unless it is a generalized tonic-clonic

seizure or status epilepticus,<sup>13</sup> the features of which include loss of consciousness. Based on the fact that the patient was able to respond to his mother during the hypertonia stage, the possibility of generalized tonic-clonic seizure or status epilepticus was unlikely. In addition, no tonic-clonic movements were observed. The patient had no history of previous seizure. The authors believe that the cause of rhabdomyolysis in this patient was hypertonia. The trigger factors were postoperative physical discomfort and insufficient pain control. Nevertheless, the possibility of seizure-induced rhabdomyolysis could not be completely excluded because simultaneous EEG was not available.

There are few studies that have reported on the association between CP and rhabdomyolysis. Intrathecal baclofen pump withdrawal, malignant hyperthermia, and repeated violent ballism are reported as causes of rhabdomyolysis in children with CP.<sup>14,15</sup> The cause of rhabdomyolysis in this case was different from those mentioned above; however, the trend of CK elevation and decline in this case was similar to that reported in the literature.<sup>1,8</sup>

A child with CP may also have nonmotor impairments, such as communication difficulty, intellectual dysfunction, learning disabilities, and sensory impairment.<sup>16</sup> As a result, the symptoms of muscle pain and weakness are easily overlooked. Fortunately, in our patient, rhabdomyolysis was diagnosed via tea-colored urine and elevated CK level, and ARF was averted by the early diagnosis, vigorous hydration and sedation.

The main goal in the treatment of rhabdomyolysis is to recognize and treat complications. Closely monitoring limb swelling, urine output, electrolytes, renal and hepatic function, and coagulation function is important. Life-threatening electrolyte imbalance should be corrected and the pressured muscle compartment should be released immediately. The pillar of treatment for rhabdomyolysis is aggressive intravascular volume supplement to promote diuresis and to dilute the toxic products.<sup>1,17</sup> Alkalinization of urine appears to be an effective method to prevent ARF.<sup>18</sup> In addition, intravenous administration of sodium bicarbonate can correct the hyperkalemia and metabolic acidosis. However, the clinical benefits of alkalinization have not been confirmed.<sup>17</sup> The roles of mannitol and furosemide in nontraumatic rhabdomyolysis are controversial. Emergent hemodialysis may be required to treat severe electrolyte imbalances, refractory acidosis and ARF.<sup>1,2,17,18</sup>

The prompt recognition of rhabdomyolysis in CP patients after surgery depends on a high level of suspicion. Routine checks of urine color after surgery is mandatory. For patients with high muscle tone, monitoring of muscle enzymes is recommended. Adequate

sedation, pain control and hydration are essential to prevent the progression of this life-threatening condition. If benzodiazepines and analgesics do not work, general anesthesia and muscle relaxant under full monitoring should be considered to stop the potential harmful abnormal muscle activity.

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