

trial. Some patients reported that they got a younger relative to call, or that they used a relative's touch-tone telephone. Some patients who were unsuccessful in accessing their messages called one of the nurses to do so; even that technique saved physician time. We learned that patients needed more warning that dictations might be delayed at times because of laboratory or physician delay. For the small number of patients who will not be happy without a written report or a personal conversation, physicians can provide that service and might be justified in charging for these personal conferences.

Most of the patients who used the message line found it easy to use and were satisfied with it. We encountered no confidentiality problems, and expect none, because patients have control of their access numbers and we use only first names in our messages. We believe our system is more confidential than sending letters or e-mail messages, or calling patients at their place of employment.

REFERENCES

1. Boohaker EA, Ward RE, Uman JE, McCarthy BD. Patient notification and follow-up of abnormal test results. *Arch Intern Med.* 1996; 156:327–331.
2. Schofield MJ, Sanson-Fisher R, Halpin S, Redman S. Notification and follow-up of Pap test results: current practice and women's preferences. *Prevent Med.* 1994;23:276–283.
3. Lind SE, Kopans D, Good MJ. Patient's preferences for learning the results of mammographic examinations. *Breast Cancer Res Treat.* 1992;23:223–232.

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Supported by a grant from the Department of Medicine, James H. Quillen College of Medicine, East Tennessee State University, Kingsport, Tennessee.

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Manuscript submitted February 2, 1999, and accepted in revised form January 10, 2000.

Focal Exertional Rhabdomyolysis Associated with a Hemangioma Steal Syndrome

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Strenuous physical activity leads to anaerobic glycolysis, with a resultant rise in blood lactic acid levels and the development of metabolic acidosis (1). Rhabdomyolysis results from skeletal muscle injury with release of



Figure 1. A large cutaneous hemangioma of the left arm, part of the left neck, and the upper left chest.

muscle cell contents (2), and may occur following exertion when energy supplies to muscle are insufficient to meet demands (2). A large cutaneous hemangioma, which can function as an arteriovenous shunt (3), may cause tissue ischemia and cellular damage through a “steal phenomenon” (4). We report a patient with a local giant cutaneous hemangioma who developed rhabdomyolysis of his left upper arm—diagnosed by a swollen extremity, elevated serum creatine kinase and aldolase levels, and magnetic resonance imaging—after intensive weight lifting. We performed serial measurements of the degree of ischemia and muscle damage during a supervised weight-lifting session, and discuss the possible pathophysiologic relation between the hemangioma and rhabdomyolysis.

CASE REPORT

A 19-year-old right-handed man experienced left upper limb pain and swelling after he began weight lifting. In five 45-minute sessions during a 10-day period, he increased the load from 15 to 30 kg in each arm. Two days after his fifth session, he was seen in the emergency department. He was afebrile, normotensive, and appeared well hydrated. The diameter of his left upper arm was 36 cm, the diameter of his left forearm was 27 cm, the diameter of his right upper arm was 29 cm, and the diameter of his right forearm was 22 cm. He had a large cutaneous hemangioma that extended from the outer third of his left hand to his neck and upper chest (Figure 1), which was in the same location as his symptoms. He denied alcohol or drug abuse. Serum creatine kinase, aldolase, alanine aminotransferase, aspartate aminotransferase,

Table 1. Serum Biochemical Values*

Days after Symptoms Began	Alanine Aminotransferase (0–40 U/L)	Aspartate Aminotransferase (0–37 U/L)	Creatine Kinase (0–190 U/L)	Aldolase (0–7.6 U/L)	Uric Acid (3–7 mg/dL)
2	349	246	7723	18	8.1
4	216	206	4958	14	7
6	84	67	654	5	5.2
10	42	51	128	3	4.8

* Values in parentheses indicate the normal range and units.

and uric acid levels were increased (Table 1). Red and white blood cell and platelet counts, and serum creatinine, myoglobin, glucose, and electrolyte levels were normal. A Doppler ultrasound of the left arm did not show deep vein thrombosis. The patient was told to refrain from physical exertion and to drink fluids. Two days later the pain and swelling persisted, although decreased in intensity; laboratory values remained abnormal (Table 1). A magnetic resonance imaging scan of his extremities was consistent with rhabdomyolysis in several muscles of the left arm and hemithorax (Figures 2 and 3). The patient's symptoms improved, and his laboratory values were normal at 10 days (Table 1), at which time his left upper arm diameter was 27 cm and his left forearm diameter was 19 cm. One month later, he was invited by the medical staff to reproduce a weight-lifting session at a lower load (3 kg in each arm) and for a shorter period of time (15 minutes), which were thought to be safe. Serum aldolase, creatine kinase, lactic acid pH, and venous blood gas values were measured before and after exercise in each arm (Table 2). When the session finished, the patient was asymptomatic. He was advised to avoid weight lifting in the future.

DISCUSSION

Severe exercise, which leads to anaerobic glycolysis due to tissue hypoxia, is characterized by lactic acid overproduction and may even cause rhabdomyolysis (1, 2, 5–9).

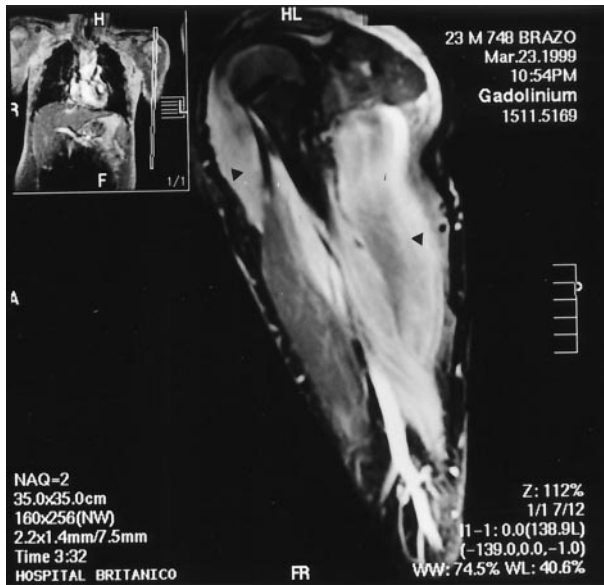
However, the physical findings in our patient, coupled with the magnetic resonance results (Figure 2) and the serum measurements (Table 2), suggest that only the left upper arm, an area dominated by a giant cutaneous hemangioma (Figure 1), was affected by rhabdomyolysis. We believe that this shunt could have stolen blood-flow under a situation of increased demand, worsening the hypoxic state and precipitating focal rhabdomyolysis. Consistent with this hypothesis, serum lactate levels were similar in both arms before physical activity, but higher in the left arm after exertion (Table 2). Metabolic acidosis and increases in arterial lactate levels do not occur until the energy requirement is about four times basal requirements (10). Oxygen levels, however, were lower in the left arm during the basal period but higher immediately after exercise. Venous oxyhemoglobin saturation was measured in the limb as a whole, and we were unable to distinguish oxygen levels in the hemangioma and in the muscle separately.

Lactate can accumulate when there is increased lactate production, diminished lactate utilization, or both. The former can occur by enhanced pyruvate production, reduced pyruvate utilization, or, as in our patient, an altered redox state in which pyruvate is converted into lactate (11, 12). Under anaerobic conditions, adenosine triphosphate (ATP) depletion may lead to the opening of potassium channels, resulting in the outflow of potassium from cells, depolarization of vascular smooth muscle cells, and decreased entry of calcium into cells, causing vasodilatation (13). Most cases of lactic acidosis are due

Table 2. Laboratory Measurements in Both Arms at Baseline, during, and after Physical Activity*

Minutes	Lactate (6–22 mg/dl)	Aldolase (0–3 U/L)	Creatine Kinase (0–190 U/L)	Venous pH (7.35–7.45)	Venous Partial Pressure of Oxygen (mm Hg)	Venous Bicarbonate (mEq/L)	Venous Oxyhemoglobin Saturation (%)
Basal	15, 16	3, 3	51, 51	7.32, 7.32	24, 20	25, 25	43, 32
2	77, 122	3, 5	50, 53	7.28, 7.22	37, 47	22, 17	63, 76
3	51, 92	3, 6	54, 60	7.30, 7.20	36, 42	23, 16	56, 73
15	50, 56	3, 5	55, 76	7.31, 7.23	32, 37	24, 19	51, 66
30	44, 46	3, 5	54, 62	7.32, 7.26	30, 30	26, 22	48, 57
80	37, 44	3, 3	59, 55	7.32, 7.32	26, 21	26, 25	44, 32

* Pairs represent values in right arm, left (affected) arm.



A



B

Figure 2. Magnetic resonance image of the left upper arm in T1 SE sequence with fat saturation showing abnormal hyperintensities of rhabdomyolyzed muscles: Major and minor pectoralis (small black head arrows), long head of triceps (large black head arrow), and short head of biceps (white head arrow).

to tissue hypoperfusion (11, 12, 14). We believe that the hemangioma led to local hypoperfusion of muscles and subsequent hypoxia which, under sustained physical activity, caused an increase in lactate production and precipitated rhabdomyolysis.

Normal subjects can develop rhabdomyolysis after seizures or intense exercise such as weight lifting (2, 5, 9, 15). Sarcolemmal damage due to exertion can interfere with

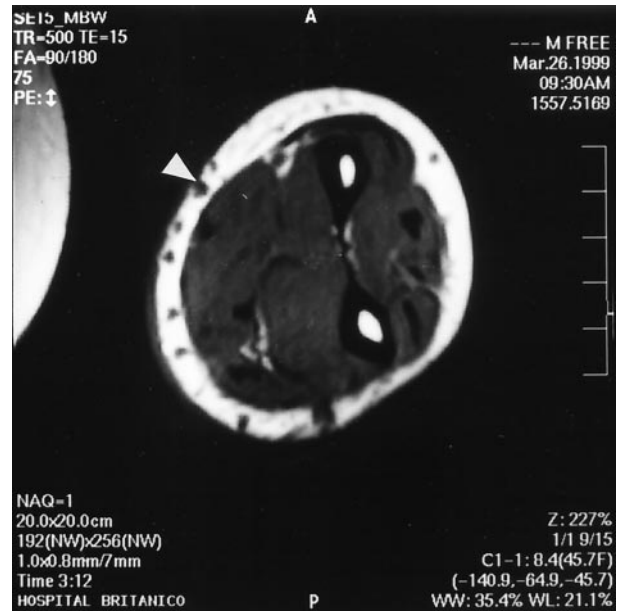


Figure 3. Magnetic resonance image in T1 SE axial sequence of left forearm showing multiple tubular structures (hemangioma) in soft tissue (head arrows).

sarcolemmal integrity and cause a leak of intracellular components (myoglobin, creatine kinase) into the plasma (16–19). Exhaustive exercise also depletes energy stores and disrupts cellular transport, raising intracellular calcium, which activates proteolytic enzymes resulting in mitochondrial damage and diminishing ATP generation. The end result is cellular anoxia and cell death (20).

Creatine kinase is a sensitive marker of muscle injury (21), whereas an increased serum myoglobin concentration and myoglobinuria are less sensitive tests (9, 22). Serum myoglobin levels may fall to normal by the time a patient is hospitalized owing to its rapid clearance from the plasma within 1 to 6 hours (2). Thus, it is not unusual for the levels of serum creatine kinase, which is cleared more slowly, to remain elevated in the absence of increased myoglobin levels (23, 24), as occurred in this patient.

The prognosis of patients with adequately treated rhabdomyolysis is generally excellent, because muscle repair mechanisms are efficient (25). Acute renal failure, one of the most serious complications, can be managed with hyperhydration, alkalinization of urine, avoidance of nephrotoxins, and if required, transient hemodialysis until renal failure resolves (2, 26–28).

Magnetic resonance imaging has a higher sensitivity for the detection of abnormal muscles (100%) than computed tomography (62%) or ultrasound (42%) (29). In magnetic resonance studies, affected muscles have an increased intensity in T2-weighted images (edema), due to inflammation and muscle necrosis (30).

Physicians should maintain a high index of suspicion for acute exertional rhabdomyolysis in patients who present with symptoms of an overexertion injury, such as pain and swelling (5). We believe vascular malformations should be considered as triggering or predisposing factors for focal rhabdomyolysis when there is muscular hyperactivity.

REFERENCES

- Shapiro J, Kaehny W. Pathogenesis and management of metabolic acidosis and alkalosis. In: Schrier R, ed. *Renal and Electrolyte Disorders*. 4th ed. Boston: Little, Brown and Company; 1992:182–184.
- Gabow P, Kaehny W, Kelleher S. The spectrum of rhabdomyolysis. *Medicine*. 1982;61:141–152.
- Enzinger F, Weiss S. *Soft Tissue Tumors*. St. Louis: CV Mosby; 1983: 379–501.
- Owen C, Mubarak S, Hargens A, et al. Intramural pressures with limb compression. Clarification of the pathogenesis of drug-induced muscle-compartment syndrome. *NEJM*. 1979;300:1169–1175.
- Line R, Rust G. Acute exertional rhabdomyolysis. *Am Fam Phys*. 1995;52:502–506.
- Schiff H, Mac Searraigh E, Kallmeyer J. Myoglobinuria, rhabdomyolysis and marathon running. *Q J Med*. 1978;188:463–472.
- Anderson J, Sander C, Larson P. Exercise myoglobinuria. *NEJM*. 1974;290:632.
- Bank W. Myoglobinuria in marathon runners: possible relationship to carbohydrate and lipid metabolism. *Ann N Y Acad Sci*. 1977; 301:942–951.
- Demos M, Gitin E, Kagen L. Exercise myoglobinemia and acute exertional rhabdomyolysis. *Arch Intern Med*. 1974;134:669–682.
- Naimark A, Wasserman K, McLroy M. Continuous measurement of ventilatory exchange ratio during exercise. *J Appl Physiol*. 1964;19: 644–652.
- Kreisberg R. Lactate homeostasis and lactic acidosis. *Ann Intern Med*. 1980;92:227–236.
- Madias N. Lactic acidosis. *Kidney Int*. 1986;29:752–763.
- Landry D, Oliver J. The ATP-ase sensitive K⁺ channel mediates hypotension in endotoxemia and hypoxic lactic acidosis in dogs. *J Clin Invest*. 1992;89:2071–2084.
- Weil M, Afifi A. Experimental and clinical studies on lactate and pyruvate as indicators of the severity of acute circulatory failure (shock). *Circulation*. 1970;41:989–1001.
- Bolgiano E. Acute rhabdomyolysis due to body building exercise. Report of a case. *J Sports Med Phys Fitness*. 1994;34:76–78.
- Better O, Abassi Z, Rubinstein I, et al. The mechanism of muscle injury in the crush syndrome: ischemic versus pressure-stretch myopathy. *Miner Electrolyte Metab*. 1990;16:181–184.
- Zager R. Rhabdomyolysis and myohemoglobinuric acute renal failure. *Kidney Int*. 1996;49:314–326.
- Heppenstall B, Scott R, Sapega A, et al. A comparative study of the tolerance of skeletal muscle to ischemia. *J Bone Joint Surg*. 1986; 68A:820–828.
- Knochel J. Rhabdomyolysis and myoglobinuria. *Semin Nephrol*. 1981;1:75–85.
- Abassi Z, Hoffman A, Better O. Acute renal failure complicating muscle crush injury. *Semin Nephrol*. 1998;18:558–565.
- Hess J, MacDonald R, Frederick R, et al. Serum creatine phosphokinase (CPK) activity in disorders of heart and skeletal muscle. *Ann Intern Med*. 1964;61:1015–1023.
- Knochel J. Rhabdomyolysis and myoglobinuria. In: Suki W, Eknoyan G, eds. *The Kidney in Systemic Disease*. 2nd ed. New York: John Wiley & Sons; 1981:263.
- Koskelo P, Kekki M, Wager O. Kinetic behavior of I-labelled myoglobin in human beings. *Clin Chim Acta*. 1967;17:339–347.
- Stone M, Willerson J, Gomez-Sanchez C, Waterman M. Radioimmunoassay of myoglobin in human serum. Results in patients with acute myocardial infarction. *J Clin Invest*. 1975;56:1334–1339.
- Poels P, Gabreels F. Rhabdomyolysis: a review of the literature. *Clin Neurol Neurosurg*. 1993;95:175–192.
- Eneas J, Schonfeld P, Humphreys M. The effect of infusion of mannitol-sodium bicarbonate on the clinical course of myoglobinuria. *Arch Intern Med*. 1979;139:801–807.
- Better O, Stein J. Early management of shock and prophylaxis of acute renal failure in traumatic rhabdomyolysis. *NEJM*. 1990;322: 825–831.
- Ron D, Taitelman U, Michaelson M. Prevention of acute renal failure in traumatic rhabdomyolysis. *Arch Intern Med*. 1984;144:277–283.
- Lamminen A, Hekali P, Tiula E, et al. Acute rhabdomyolysis: evaluation with magnetic resonance imaging compared with computed tomography and ultrasonography. *Br J Radiol*. 1989;62:326–330.
- Revelon G, Rahmouni A. Acute swelling of the limbs: magnetic resonance pictorial review of fascial and muscle signal changes. *Eur J Radiol*. 1999;30:11–21.

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Manuscript submitted September 28, 1999, and accepted in revised form January 7, 2000.

The R131 Low-affinity Allele of the Fc Gamma RIIA Receptor Is Associated with Systemic Lupus Erythematosus but Not with Other Autoimmune Diseases in French Caucasians

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The evidence that there is a genetic susceptibility to systemic lupus erythematosus (SLE) in humans is based on its high concordance rate (29% to 57%) in identical twins and on the relatively high incidence (10% to 12%) of familial cases (1). Since the production