CASE REPORT

Life-threatening rhabdomyolysis after prolonged hypoperfusion of the lower legs

Sebastian Ørskov Abrahamsen,¹ Maiken Stilling²¹Department of Orthopedic, Regionshospitalet Randers, Randers, Denmark²Department of Orthopedic, Aarhus University Hospital, Aarhus, Denmark**Correspondence to**

Dr Sebastian Ørskov Abrahamsen, sebastianabrahamsen@hotmail.com

SUMMARY

Rhabdomyolysis is a potentially life-threatening condition to be considered when muscular damage is suspected. We present a case of a 39-year-old woman who developed rhabdomyolysis after prolonged circulatory insufficiency to the legs, leading to life-threatening complications including bilateral tibial compartment syndrome, acute kidney failure, hyperkalaemia with cardiac arrhythmia and finally lifesaving bilateral transfemoral amputation. This case emphasises that early diagnosis, removal of triggering factor, early and aggressive fluid therapy and treatment and prevention of complications are essential for the patient's morbidity and mortality.

BACKGROUND

This case is interesting primarily because it confirms the importance of suspecting rhabdomyolysis (RM) even in minor or non-traumatic cases, second because there have only been a few cases reported regarding non traumatic bilateral compartment syndrome of the tibias.¹ There is no consensus regarding the recommendations of early fluid therapy before the chemical markers confirm the diagnosis, but it is possible that early aggressive therapy could have changed the traumatising outcome for this patient. More clinical research is needed and recommended.^{2 3}

CASE PRESENTATION

A 39-year-old, moderately obese woman was brought into the emergency department of a smaller Danish hospital complaining of mild diffuse lower leg pain and the lack of sensibility below knee level. She had been kneeling in a praying position for 8 h after she fell out of her bed at night because she, as explained, was too tired to change her position. There was no history of injury. The patient had previous episodes of alcohol and substance abuse, primary benzodiazepines. She had no other comorbidities, was not taking any regular medications including oral contraceptives and was a non-smoker.

The objective findings were minor redness and a few bullae located at both knee caps and the lateral side of both tibias, slightly decreased active movement of the knee joints and no active movement from the ankle joint and also distally. Initially, there were no clinical signs of oedema or increased pressure of the tibial compartments, except from the earlier mentioned symptoms. X-ray examinations of both knees were normal. Biochemical analysis showed compensated metabolic acidosis, slightly

increased Creatine Kinase MB Isoenzyme (173.4 µg/l, ref. <4.0 µg/l), C reactive protein (CRP) (46.8 mg/l, ref. <8.0 mg/l) and leucocytes (27.1 times 10⁹/l, ref. 3.5–10.0 times 10⁹/l).

On the suspicion of erysipelas, the patient was transferred to a medical department, when a myoglobin above 30 000 µg/l (ref. 25–58 µg/l) and (no initial creatine kinase was taken) creatine kinase above 20 000 U/l (ref. 50–150 U/l) were measured, and as a result the patient was transferred to the intensive care unit (ICU) for forced diuresis, sedation and intubation. Ten hours after hospital admission, the patient developed bilateral tensed and swollen compartments of the tibia, and on the suspicion of acute compartment syndrome, acute bilaterally fasciotomy was performed. The muscle tissue was grey and without any reaction to electrostimulation. Fourteen hours later, the patient developed anuria and azotaemia, and dialysis (continuous venovenous hemodiafiltration) was started. The patient slowly recovered clinically, and the blood tests slowly declined (CRP: 36.0 mg/l, myoglobin: 868 µg/l, creatine kinase: 1226 U/l, creatine: 144 µmol/l (ref. 45–90 µmol/l) and glomerular flow rate: 36 ml/min (ref. >60 ml/min)). However, in the following days, repeated debridement of the non-vital muscles in the crural compartments was necessary.

At day 12, myoglobin (1014 µg/l), creatine kinase (665 U/l) and potassium (5.9 mmol/l, ref. 3.6–5.0 mmol/l) increased again, and most of the muscle tissue on both tibias was judged non-vital. The patient was recommended bilateral transfemoral amputation, but refused. Over the next few days, the biochemical and the clinical condition of the patient declined, and on day 16 she was in toxic shock (GCS=9), tachycardia (pulse 152), low blood pressure (98/71) with cardiac arrhythmias and was febrile (temperature 40.2°C). She had several episodes of convulsions treated with intravenous diazepam. The myoglobin (4467 µg/l), creatine kinase (394 U/l) and potassium (6.1 mmol/l) levels elevated over time.

Senior surgeons now consensually agreed that transfemoral amputation would be the only chance to save the patient's life, and her close relatives consented on behalf of the patient.

Postoperatively, the patient improved daily, and myoglobin and creatine kinase levels decreased slowly. Finally, on day 24, the patient was transferred from the orthopaedic department to the nephrologists because of lasting anuria.

In the months after surgery, the patient's kidney function improved, and now 1.5 years after surgery

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she has normal kidney function, and dialysis is no longer indicated. In the weeks after the surgery, a revision of the limbs, because of infection and not vital tissues, was performed with a good result, and healing of the cicatrices occurred after approximately 4 weeks postoperation. She is still treated with strong painkillers (opiates), non-steroidal anti-inflammatory drugs and antiepileptic drugs because of phantom limb pains.

Because of moderate obesity, she is not a candidate for transfemoral prosthesis and she is getting around using her wheelchair. By her own words, she is doing better, and she has a positive perspective on life though she is struggling with psychological problems primarily because of her limited mobility.

INVESTIGATIONS

It is well known that the causes of rhabdomyolysis are multifactorial and the diagnosis must always be kept in mind when muscular damage is suspected.^{3–5} An elevated plasma creatine kinase level is the most sensitive parameter pertaining to muscle injury.² Values 5–10 times greater than normal is diagnostic for RM.²

Once the diagnosis of RM is established, a search must be instituted for a cause. A careful history and physical examination may reveal the underlying aetiology of RM or at least may help in the selection of the appropriate diagnostic workup. However, in many cases the history and clinical examination are not conclusive and the nature of the investigations depends on the disorder suspected, for example, measuring of the compartment pressure in the tibias could be performed when compartment syndrome is suspected, as with the presented case, even though the diagnosis, compartment syndrome, most often is based on the clinical findings. Toxicological screens should be performed if drugs are a suspected causal agent, whereas appropriate cultures and serological studies should be performed if infections are suspected. Endocrine assay and blood chemistry may be necessary to confirm the suspected endocrine and metabolic disorders. Rarely genetic analysis and muscle biopsy may be indicated in patients with suspected genetic disorders.^{1–3}

Arterial blood gas analysis is a useful tool to evaluate acid-base balance, whereas ECG is helpful to evaluate for cardiac arrhythmias related to hyperkalaemia or hypocalcaemia. Standard blood samples, screening the different organ systems including complete blood count, should be performed.^{2,3}

Acute renal failure, as a consequence of precipitation of the tubules with toxic myoglobin, is seen in 10–50% of cases.^{3,4}

DIFFERENTIAL DIAGNOSIS

As mentioned earlier, any damage to the myocyte can result in RM, and therefore the aetiological spectrum of RM is extensive. The most common causes of RM in adults are illicit drugs, alcohol abuse, medical drugs, muscle diseases, trauma, seizures, excessive muscular activity, muscle infections and immobility. Genetic disorders such as McArdle's disease and Duchenne's muscular dystrophy are relatively rare causes of RM, but must be kept in mind, especially with a history of recurrent episodes of RM and with a family history of muscular disease. In many cases, the aetiology of RM cannot be identified.^{2–5}

Complications of RM include hypovolaemia—because of the influx of extracellular fluid into the necrotic muscle cell; compartment syndrome—because the ischaemic and oedematous muscle further raises intracompartmental pressure potentiating a vicious cycle of continuing ischaemia; arrhythmias and cardiac arrest—because of hyperkalaemia and hypocalcaemia, disseminated intravascular coagulation which probably results from the activating of the clotting cascade by components released from

the damaged muscle cells, hepatic dysfunction, acidosis and acute renal failure. Any disease presenting some of the earlier mentioned symptoms or organo physiological manifestations may be misdiagnosed as rhabdomyolysis. The differential diagnoses to rhabdomyolysis are therefore many. When muscular damage is suspected, an elevated plasma creatine kinase level will give the correct diagnosis.^{1–5}

TREATMENT

No randomised controlled trials of treatment have been conducted and most of the evidence is based on retrospective clinical studies, case reports and animal models.² There is a clear consensus that early diagnosis, elimination of the cause to RM, early aggressive fluid therapy and treatment and prophylaxis of complications are decisive for the patients morbidity and mortality,^{2–5} which this case emphasises.

OUTCOME AND FOLLOW-UP

The prognosis of RM is heavily dependent upon the underlying aetiology and the associated comorbidities. The evidence from case reports and small retrospective studies suggests that RM and the renal function, when treated early and aggressively, have excellent prognosis.

The need for follow-up depends on the aetiology and associated comorbidities, but, most importantly, there should be a close follow-up on the kidney function.

DISCUSSION

RM is a potentially life-threatening condition that can develop after any damage to the myocyte leading to the release of muscle enzymes, myoglobin, potassium, calcium and other intracellular components into the bloodstream.^{2,4,5} The classic findings of muscular aches, weakness and tea-coloured urine are non-specific and may not always be present,² which is why the diagnosis should always be considered with suspicion of muscular damage.^{3,4,5} Traumatic RM is the most common presentation, but even non-traumatic RM may be life-threatening as with the presented case.

We report a case where the patient developed atraumatic bilateral tibial compartment syndrome leading to massive RM after prolonged circulatory insufficiency of the legs, leading to life-threatening complications, including bilateral tibial compartment syndrome, acute kidney failure, hyperkalaemia with cardiac arrhythmia and finally necessitating bilateral lifesaving transfemoral amputation.

Bilateral compartment syndrome of the legs is a rare presentation with potentially catastrophic complications. Most cases have a clear aetiology based on the patient's history as with the presented case.¹ Khan *et al.* summarised some of the reported cases of atraumatic bilateral compartment syndrome, from 1999 to 2009 (eight case reports), and found that there have been a number relating to limb position, as with the presented case: prolonged kneeling position during spinal surgery,⁶ horse riding⁷ and prolonged lipotomy position.⁸ Other causes include excessive exercise and medical or alcohol-induced bilateral compartment syndrome.¹

In the published reports of acute atraumatic bilateral compartment syndrome, the diagnosis has often been delayed, leading to negative sequelae.¹ In the case presented, blood results and a high index of suspicion for compartment syndrome allowed early diagnosis. Furthermore, successful treatment of this patient's condition was possible due to a well-coordinated multidisciplinary team approach. Early involvement of physicians, surgeons and intensive care teams prevented further insult to

the renal system, and together with bilateral trans-femoral amputation saved the patient's life.

Finally, appropriate input from tertiary centres with a specialist renal unit, orthopaedics and physiotherapists enabled optimal functional recovery.

Learning points

- ▶ Rhabdomyolysis (RM) should always be considered with suspicion of muscular damage.
- ▶ An elevated plasma creatine kinase level is the most sensitive parameter pertaining to muscle injury.
- ▶ Elimination of the cause to RM, early aggressive fluid therapy and treatment and prophylaxis of complications are decisive for the patient's morbidity and mortality.

Contributors SØA treated the patient discussed in this case report and he also wrote the case report and collected all the necessary data about the patient and about the topic. MS gave feedback and comments in the revising of this paper.

Competing interests None.

Patient consent Obtained.

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