Fukushima J. Med. Sci.,
Vol. 56, No. 2, 2010

[Case Report]

TWO CASES OF ACUTE ATRAUMATIC COMPARTMENT SYNDROME COMPLICATED WITH SEVERE HEAT STROKE

YUKIHIRO IKEGAMI, ARIFUMI HASEGAWA, YASUHIKO TSUKADA, YOSHINOBU ABE, JIRO SHIMADA and CHOICHIRO TASE

Department of Emergency and Critical Care Medicine, Fukushima Medical University School of Medicine, Fukushima, Japan

(Received March 25, 2010, accepted July 17, 2010)

Abstract : Acute compartment syndrome is a life-threatening complication in trauma patients. Not only regional neuromuscular disability, but also systemic organ disorders can result from prolonged tissue ischemia. In this report, we describe 2 cases of acute atraumatic compartment syndrome complicated with severe heat stroke. In both cases, emergency fasciotomy was rapidly performed after recognition of the syndrome, but serious regional neuromuscular disabilities remained. Microvascular endothelial injury is an important mechanism of acute atraumatic compartment syndrome. Thrombi diffusely formed in the compartmental space hinder establishment of reperfusion even after fasciotomy. Furthermore, disruption of fibrinolysis due to heat stroke could enhance this damage.

Key words : atraumatic compartment syndrome, fasciotomy, heat stroke, thrombolysis

INTRODUCTION

Acute compartment syndrome is characterized by high pressure in compartmental spaces[1,2]. This high pressure causes serious muscle ischemia in compartmental spaces. If tissue hypoxia is prolonged, not only regional neuromuscular disability, but also systemic organ disorders may occur. In cases of delayed treatment, patients suffer from severe sequelae. Clinically, acute compartment syndrome is classified into 2 types based on developmental mechanisms. Most cases are caused by limb traumas including open or closed fractures[3], and rare cases are caused by atraumatic mechanisms[4].

In severe acute compartment syndrome, it is necessary to rapidly attempt to decompress intracompartmental pressure by emergency fasciotomy to restore regional neuromuscular function. Generally, fasciotomy performed by an orthopedic doctor is very effective. However, in some cases of acute atraumatic compartment syndrome, compartmental reperfusion cannot be established. Traumatic cases and atraumatic cases likely involve different developmental mechanisms. We speculate that the lack of reperfusion in acute atraumatic compartment syndrome is caused by systemic microvascular endothelial injury.

In this report, we describe 2 cases of acute atraumatic compartment syndrome. In both cases, very rare atraumatic mechanisms caused serious compartmental ischemia of the lower limbs. Although emergency fasciotomy was performed, serious regional neuromuscular disabilities were irreversible.

CASE REPORT

CASE 1

A 63-year-old-man with severe dementia due to Pick's disease exited his home in the middle of summer and did not return, and he was found 20 hours later. When he was found, he was sitting on

池上之浩，長谷川有史，塚田泰彦，阿部良伸，島田二郎，田勢長一郎
Corresponding author : Yukihiro Ikegami E-mail address : yikegami@fmu.ac.jp
http://www.jstage.jst.go.jp/browse/fms http://fmu.ac.jp/home/lib/F-igaku/

the ground in Japanese fashion (kneeling with buttocks resting on heels). We suspected that he had severe heat stroke because he was exposed to hot and humid conditions for a long period of time. His rectal temperature was over 40 degrees centigrade upon hospital admission. Laboratory findings showed remarkably high serum creatinine kinase levels (24,825 U/L), which further increased to 81,300 U/L 3 hours after hospitalization. There was no metabolic acidosis but severe hypovolemia was suspected (total protein : 8.8 g/dl ; hematocrit : 55%). We observed extended vesiculation, and swelling and tenderness on the patient's bilateral lower limbs (Fig. 1). There was no bone fracture on X-ray examination. When asked, the patient confessed that he had remained in the same position (Japanese fashion) for 8 hours. We suspected development of acute compartment syndrome due to atraumatic mechanisms. His right anterior tibial compartment pressure was 85 mmHg and left anterior tibial compartment pressure was 75 mmHg as measured by Intra-Compartmental Pressure Monitor System (Stryker® Surgical, Kalamazoo, USA). We immediately performed emergency fasciotomy (anterior lateral and posterior medial incision) in both limbs and compartmental pressure was decompressed under 20 mmHg. After fasciotomy, anterior tibial compartmental pressures in both limbs were maintained under 20 mmHg. However, the patient's compartmental blood circulation did not recover and muscle necrosis of both lower limbs irreversibly developed. Although we performed debridement of the focal infectious muscle on the

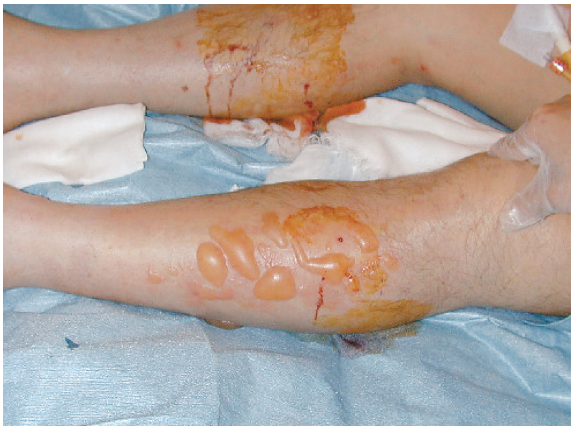

Fig. 1. Lower legs of case 1. Extended vesiculation, marked swelling, and tenderness on both lower legs.

Fig. 2. The deficit of tissue in the left lower leg was protected by a free flap using the latissimus dorsi on the 59th hospital day.

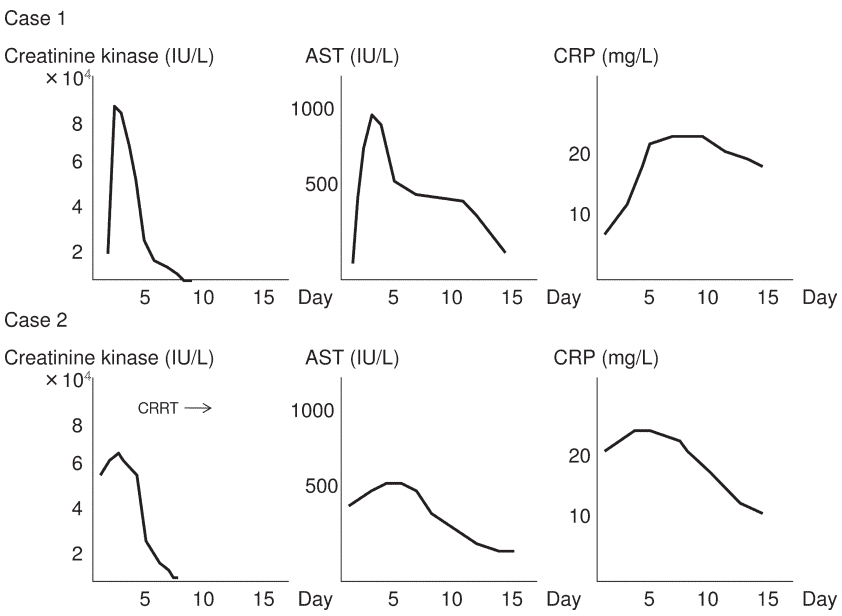

Fig. 3. Changes of clinical data in Case 1 and Case 2

18th and 38th day, the patient did not recover from the infection. On the 59th day, the deficit of tissue in the left lower limb was protected by a free flap using the latissimus dorsi (Fig 2.). Even after this radical operation, it was necessary to repeat the debridement for intractable focal infection of the left lower limb. We performed this surgical procedure 4 times in 90 days. Finally, the patient was saved but suffered from severe muscular dysfunctions of both lower limbs. The changes in laboratory findings are shown in Fig. 3.

CASE 2

A 64-year-old man lost consciousness in a steam bath in a hotel after excessive alcohol intake. The hotel officer found him lying on the floor of a steam bath in the left lateral position after the alcohol intake. He was transported to our hospital by ambulance. In the emergency room, his rectal temperature was over 41 degrees centigrade. We diagnosed him with severe heat stroke, and rapid fluid resuscitation and whole body cooling were performed. Laboratory findings showed very high serum creatinine kinase levels (58,250 U/L) on admission. Metabolic acidosis developed (pH : 7.22) and concentration of the blood (total protein : 9.0 g/dl ; hematocrit : 55%) due to severe hypovolemia. In the emergency room, the patient complained of dull pain from the left hip to the left lower limb. Swelling, tenderness, and vesiculation developed progressively after arrival, and we suspected complications of acute compartment syndrome. Three hours after hospitalization, the patient's left anterior tibial compartment pressure was over 60 mmHg as measured by Intra-Compartmental Pressure Monitor System (Stryker® Surgical, Kalamazoo, USA). We immediately performed emergency fasciotomy (anterior lateral and posterior medial incision) and compartmental pressure was decompressed under 20 mmHg. After fasciotomy, anterior tibial compartmental pressures were maintained under 20 mmHg and serum creatinine kinase levels had decreased after several days. However, the patient's compartmental blood circulation did not recover and broad muscle necrosis developed. While he was treated daily by orthopedic specialists, the necrotic area progressively increased. We decided that retaining the lower limb was not possible and his left lower limb was amputated on the 10th day. The changes in laboratory findings are shown in Fig. 3.

DISCUSSION

In our 2 cases, acute compartment syndrome developed without traumatic episodes. Atraumatic compartment syndrome is very rare, and there have been only a few reports associated with drug abuse or surgical position[5,6]. Generally, the extended immobile positioning due to deep coma is an important factor in the development of atraumatic compartment syndrome. However, our first case represented a very rare case because extended bilateral atraumatic compartment syndrome developed without a coma. Extensive muscle necrosis quickly developed even though emergency fasciotomy was rapidly performed in both cases. A poor prognosis of neuromuscular function in atraumatic cases has been previously reported[4]. In addition, our patients unfortunately suffered from serious sequelae in the lower limbs.

We evaluated the mechanisms that severely affected compartmental neuromuscular function in our atraumatic cases. Generally, collapse of the microvascular circulation in the compartmental spaces slowly develops from muscle swelling in traumatic cases. Therefore, the establishment of complete ischemia took a while. On the other hand, compartmental microvascular circulation was completely collapsed by direct compression of capillary vessels in our patients. Consequently, we speculate that ischemia in our patients was prolonged.

In atraumatic cases, continuous collapse by compression directly causes capillary vessel injury of compartmental vascular endothelial cells, and several adhesive factors are released. In addition, complete elimination of shear stress in compartmental vessel walls suppresses release of tissue plasminogen activator (t-PA) by remaining in the same position. These adhesive factors and insufficiency of t-PA contribute to the diffuse formation of microthrombi in compartmental spaces.

Our patients developed complications of severe heat stroke. Severe blood adhesion due to vascular hypovolemia initially disrupted compartmental blood perfusion. Severe systemic hyperthermia causes systemic activation of vascular endothelial cells. Activated endothelial cells reduce production of thrombomodulin (TM) and also cause production of excessive amounts of plasminogen activator inhibitor 1 (PAI-1). A decrease in TM causes systemic hyper-coagulability. PAI-1 combines with free t-PA to form a t-PA/PAI-1 complex, and consumption of t-PA causes fibrinolysis[7]. Unfortunately, we did

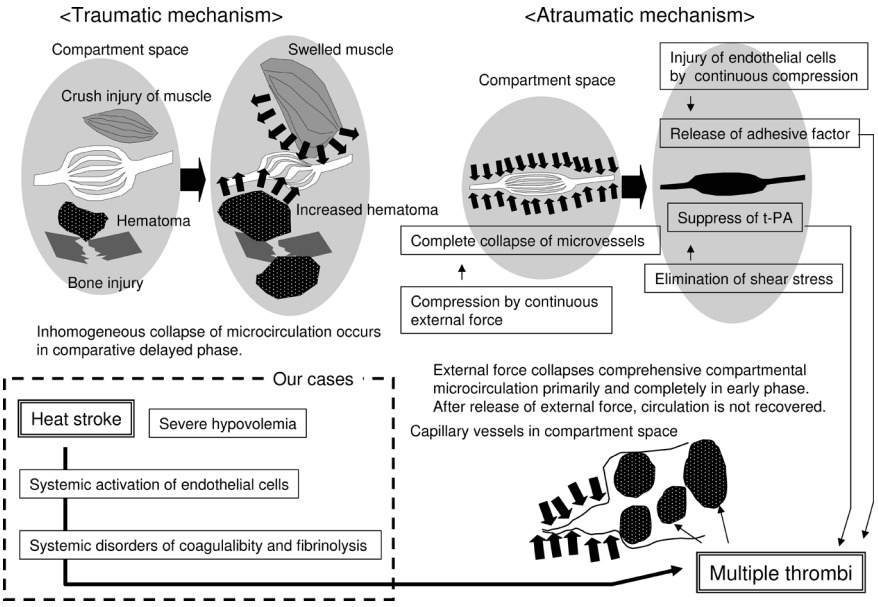

Fig. 4. Schema of mechanism

not frequently evaluate our patients' blood data, and therefore, development of disorders of coagulability and fibrinolysis cannot be proved. However, we believe that complicated heat stroke consequently caused severely affected coagulability and fibrinolysis, enhancing the development of compartmental microvascular thrombi. Even after decompression by emergency fasciotomy, we could not achieve microvascular reperfusion in compartmental spaces. The schema of the mechanism in traumatic compartment syndrome and atraumatic compartment syndrome is presented in Fig. 4.

In acute atraumatic compartment syndrome, muscle circulation in some cases cannot be sufficiently restored by fasciotomy because of disruption of coagulofibrinolytic function. If patients present with any symptoms of compartmental ischemia after fasciotomy, post-fasciotomy angiography should be performed to assess the presence of residual thrombi[8]. Mechanical thrombectomy or systematic thrombolytic therapy using t-PA is inadequate because there are risks of post-operative bleeding, and microthrombi diffusely distribute. For such cases, several reports have recommended local continuous thrombolytic therapy using urokinase[9]. Ali *et al.* recommended percutaneous isolated limb perfusion using high dose urokinase[10]. Currently, effects of platelet glycoprotein IIb/IIIa inhibitors on ischemic limbs have been reported[11].

CONCLUSION

In suspicious cases of acute atraumatic compartment syndrome, compartmental pressure should be rapidly monitored, and if necessary, emergency fasciotomy should be performed to achieve decompression. However, in some cases, sufficient compartmental reperfusion cannot be obtained by surgical decompression due to diffuse microthrombi produced by vascular endothelial injury. If patients present with any symptoms of compartmental ischemia, post-fasciotomy angiography and selective thrombolytic therapy using urokinase should be performed.

REFERENCES

1. Mubarak SJ, Owen CA, Hargens AR, Garetto LP, Akeson WH. Acute compartment syndromes : diagnosis and treatment with the aid of the wick catheter. J Bone Joint Surg, **60** : 1091-1095, 1978.
2. Allen MJ, Stirling AJ, Crawshaw CV, Barnes MR. Intracompartmental pressure monitoring of leg injuries. An aid to management. J Bone Joint Surg, **67** : 53-57, 1985.
3. Gulli B, Templeton D. Compartment syndrome of the lower extremity. Orthop Clin North Am, **25** : 677-683, 1994.
4. Ochoa-Gomez J, Villar-Arias A, Aresti I, Marco-Aguilar P. A case of severe hyperkalaemia and compartment syndrome due to rhabdomyolysis after drugs abuse. Resuscitation, **54** : 103-105, 2002.
5. Mulhall JP, Drenzner AD. Postoperative compartment syndrome and the lithotomy position : a report of these cases and analysis of potential risk

factors. Conn Med, **57** : 129–133, 1993.
6. Franc–Law JM, Rossignol M, Vernec A, Somogyi D, Shrier I. Poisoning–induced acute atraumatic compartment syndrome. Am J Emerg Med, **18** : 616–621, 2000.
7. Asakura H, Jokaji H, Saito M, Uotani C, Kumabashiri I, Morishita E, Yamazaki M, Matsuda T. Changes in plasma levels of tissue–plasminogen activator/inhibitor complex and active plasminogen activation inhibitor in patients with disseminated intravascular coagulation. Am J Hemato, **36** : 176–183, 1991.
8. Reuss PM, Rosen RJ, Adelman M. Compartment syndrome complicating lower extremity thrombolysis. J Vasc Interv Radiol, **10** : 1075–1082, 1999.
9. Tsuda M, Nakamura M, Yamada Y, Saito H, Ishibashi T, Takahashi S. Acute superior mesenteric artery embolism : rapid reperfusion with hydrodynamic thrombectomy and pharmacological thrombolysis. J Endovasc Therap, **10** : 1015–1018, 2003.
10. Ali AT, Kalapatapu VR, Bledsoe S, Moursi MM, Eidt JF. Percutaneous isolated limb perfusion with thrombolytics for severe limb ischemia. Vasc Endovascular Surg, **39** : 491–497, 2005.
11. Laird JR. The management of acute limb ischemia : techniques for dealing with thrombus. J Interv Cardiol, **14** : 539–546, 2001.