An unusual gangrene of multiple limbs due to tight ligatures in an alcoholic patient

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Citation

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Abstract

The traditional bone setter's gangrene due tight splints is a well-known entity. Gangrene usually supervenes following prolonged pressure of tight splintage by traditional bone setters in the process of treating fractures. We present an unusual case of gangrene in multiple limbs in a chronic alcoholic due to tight ligatures in a non-traumatic situation. Such a case has not been reported previously in the literature. The authors propose a combined effect of tight constricting ligatures leading to prolonged venous occlusion local tissue trauma, acute alcohol toxicity, altered sensorium, decreased tissue perfusion and peripheral neuropathy as the possible mechanism.

INTRODUCTION

Matsen [1] defined the compartment syndrome as "a condition in which increased pressure within a limited space compromises the circulation and function of the tissues within that space." Most compartment syndromes are associated with traumatic insults, but the condition also occurs after reperfusion, following a period of ischemia, burns, and prolonged limb compression after drug abuse or poor positioning during prolonged surgical procedures [2, 3]. Prolonged venous occlusion and blood dyscresias are other causative factors [4]. The compartmental perfusion pressure, which is the mean arterial pressure minus the compartment pressure, should be above 70-80 mmHg. Both increasing the compartmental pressure and decreasing the perfusion pressure can lead to a compartment syndrome [2].

The traditional bone setter's gangrene due tight splints is a well-known entity $[_5, _6]$. The present study reports gangrene in the three limbs of an alcoholic patient after tight ligatures around the limbs. The authors are not aware of any such case report to the best of their knowledge.

CASE REPORT

A 55-year-old male presented with tense swellings of bilateral feet and left hand. A detailed history revealed; patient was a chronic alcoholic since 15 years. He had an altercation with family members in an inebriated state seven days back. He was tied by the family members to the bed with the help of plastic ropes around his legs (above ankles) and around the distal forearms for whole night for about 12 hours. Patient developed painful swelling of both the feet and left hand for which he did not seek proper medical treatment except for taking self-medication. He started developing blackening of his toes of both feet and left hand on third day of the incident for which he sought treatment from a private practioner. Amputation of the great toe and debridement of the gangrenous portion was done. On seventh day, he was referred to our tertiary care centre for further management. No other positive history could be sought. On examination, sensation to light touch was decreased throughout the bilateral feet and fingers of the left hand. The swelling had decreased by this time and there were gangrenous changes of both the feet and left hand. (Fig. 1) left hand fingers had started developing contractures.

Figure 1

Figure 1: Gangrene of both feet and left hand. Right great toe amputated. Ligature marks are visible (arrows).



The patient had impression of ligatures on all the four limbs at bilateral wrists and ankles. Bilateral pedal and radial pulses were palpable. Plain radiographs of the bilateral feet and hands demonstrated no fractures. Color Doppler of the limbs for arterial and venous system did not reveal any abnormality. A coagulation panel (partial thromboplastin time, platelet count, fibrinogen level, and liver function tests) showed no abnorm`alities. Serial wound debridements were required over next 2 weeks until wounds became cleanly granulated, and delayed split-thickness skin graftings were performed.

DISCUSSION

Four-extremity gangrene or gangrene in multiple limbs is a rare phenomenon. Symmetrical peripheral gangrene may manifest in conditions associated with sepsis, low output states, vasospastic condition, myeloproliferative disorders or in hyperviscosity syndrome [7]. Hacking et al reported a quadruple amputation following a meningococcal septic shock in a patient who also suffered from cerebral and peripheral nerve dysfunction [8]. Mokoena and Hadley [9] reported multiple limb gangrene following dehydration in children. The most affected child had gangrene and mummification of all limbs as well as gangrene of the ear lobes and the tip of the nose. Dhawan and Wang [10] reported four-extremity gangrene of both hands and legs associated with crack cocaine abuse. Patient also had decreased mental responsiveness and developed bilateral hand compartment syndrome requiring emergency fasciotomy. Gutierrez et al. [11] reported cocaine-induced peripheral vascular occlusion resulting in progressive bilateral gangrene of both legs necessitating bilateral below-knee amputations.

Tight splintage in the limbs can cause gangrene necessitating amputation [$_{5,6}$]. Gangrene usually supervenes following prolonged pressure of tight splintage by traditional bone setters in the process of treating fractures [$_6$]. External compression of the lower legs (because of positioning, stirrups, or antiembolism stockings) is a risk factor for development of the compartment syndrome of the lower leg after surgery in the lithotomy position [$_{12}$]. The pathogenesis of lower limb compartment syndrome or well leg compartment syndrome comprises increase in intracompartmental pressure and hypo perfusion, resulting in ischemia and necrosis [$_{13}$].

The patient in the present case report developed compartment syndromes of bilateral feet and left hand due to tight ligatures around the limbs and subsequently gangrenous changes in the overlying skin and fascia. As the patient presented late with advanced stage of compartment syndrome with gangrene, the intra-compartmental pressures were not measured. There is a dynamic relationship between the level of the intra-compartmental pressure and the duration of elevated pressure; the longer the delay to fasciotomy, the worse the outcome [2]. If the delay is more than 12 h, bad results are inevitable [2] which is the scenario in the present case.

The clinical signs of compartment syndrome can be elicited only in the fully conscious patient. Early diagnosis is difficult in patients with CNS compromise, the very young and the very old, and in patients with substance abuse. In patients with altered consciousness due to drug or alcohol abuse, prolonged limb compression can lead to soft tissue injury and compartment syndrome [2]. 79% patients had an obtunded sensorium-either because of a serious illness or injury or secondary to prolonged anesthesia-when the compartment syndrome was recognized [3].

Pathogenesis of the compartment syndrome may be multifactorial in the present case. Schuckit MA [14], in the chapter on Alcohol and Alcoholism describes, "Ethanol is a central nervous system depressant that decreases neuronal activity, although some behavioral stimulation is observed at low blood levels. Chronic high doses cause peripheral neuropathy in 5% to 15% of alcoholics: patients experience bilateral limb numbness, tingling, and paresthesias, all of which are more pronounced distally. Acutely, ethanol decreases myocardial contractility and causes peripheral vasodilatation, with a resulting mild decrease in blood pressure. Peripheral vasodilatation and decreased blood pressure will lead to decrease in the perfusion pressure." In the present case, patient in agitated state under the influence of alcohol might had caused repeated tissue trauma at ligature sites because of multiple attempts of freeing himself and overall self neglect by the patient for not seeking proper medical care for three days might had further perpetuated the problem. We think the possible mechanism of compartment syndrome and gangrene in our case is "a combined effect of tight constricting ligatures leading to prolonged venous occlusion local tissue trauma, acute alcohol toxicity, altered sensorium, decreased tissue perfusion and peripheral neuropathy."

CONCLUSIONS

In summary, we present an unusual case of gangrene in multiple limbs in a chronic alcoholic due to tight ligatures not reported previously. The authors propose a combined effect of tight constricting ligatures leading to prolonged venous occlusion local tissue trauma, acute alcohol toxicity, altered sensorium, decreased tissue perfusion and peripheral neuropathy as the possible mechanism. There is a need to educate the society to avert such type of mishaps.

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