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A rare case of symmetrical four limb gangrene following emergency neurosurgery

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ABSTRACT

INTRODUCTION: The authors report a case of symmetrical peripheral gangrene (SPG) following emergency neurosurgery.

PRESENTATION OF CASE: A 35-year-old female presented to hospital in Thailand with nausea, headache, and subsequent seizures. She was found to have a large intracranial space-occupying lesion with mass effect. Following emergency surgical debulking and decompression, she suffered from severe sepsis with multiple organ failure, treated with high dose intravenous vasopressors and developed secondary gangrene in all four limbs. She was repatriated to the UK with a baseline GCS of 8 and multiple postoperative medical complications. With initial conservative management, the patient made a prolonged but satisfactory progression to recovery prior to semi-elective debridement and selected digit amputation of the gangrene.

DISCUSSION: This is the first reported case of four limb symmetrical peripheral gangrene following an emergency craniotomy.

CONCLUSION: Although rare, SPG is a substantial complication with high mortality and morbidity and therefore should be especially taken into account for emergency intracranial pathologies in neurosurgical patients, particularly if they require emergency surgery.

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1. Introduction

Symmetrical peripheral gangrene (SPG) is defined as distal ischaemic damage in two or more extremities, without large vessel obstruction or vasculitis [1]. It is associated with various underlying medical conditions and is a rare but significant complication of septicemia, with up to 40% mortality [2]. Aggravating factors include asplenia, immunosuppression, previous cold injury to extremities, diabetes mellitus, renal failure, increased sympathetic tone and the use of vasopressors [3–5].

We report a case of severe four limb SPG secondary to combined septicaemia and vasopressor use following emergency surgery for a haemorrhagic brain tumour in a young patient.

2. Presentation of case

A 35-year-old female presented to hospital in Thailand with nausea, headache, and subsequent seizures. An MRI head showed a left frontal lobe tumour with surrounding oedema, midline shift, uncal/transtentorial herniation, and hydrocephalus. An emergency craniotomy and debulking/decompression was performed. Following surgery, the patient developed severe urosepsis, septic shock and multi organ failure, which required treatment with high dose intravenous (IV) noradrenaline. Unfortunately, this led to secondary ischaemia in all four limbs. The histology of the tumour confirmed a WHO grade III anaplastic astrocytoma.

The patient had a previous history of an astrogloma resected 10 years ago and Crohn's disease. She was previously fit otherwise, with no predisposing factors for postoperative septicaemia.

After 5 weeks of intermittent intubation and ventilation while receiving treatment for other postoperative complications including acute respiratory distress syndrome (ARDS), disseminated intravascular coagulation (DIC), hospital acquired pneumonias and acute kidney injury (AKI), she was repatriated to the UK with a GCS of 8 (E4V1M3) and four-limb gangrene (Figs. 1–4). At this stage she was NG fed due to unsafe swallowing, unable to speak or obey commands and only moved her limbs in response to pain. On admission to the UK hospital, Hb was 89 g/L, platelets was $382 \times 10^9/L$, WBC

Abbreviations: SPG, symmetrical peripheral gangrene; GCS, Glasgow coma score; DIC, disseminated intravascular coagulation; ARDS, acute respiratory distress syndrome; AKI, acute kidney injury.

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Fig. 1. Dorsal and palmar aspects of right hand.



Fig. 2. Dorsal and palmar aspects of left hand.

was $12 \times 10^9/L$, CRP was 11 mg/L, urea was 17.6 mmol/L, creatinine was $313 \mu\text{mol/L}$. Blood, wound, faeces, and catheter tip cultures were all negative. Subsequent rectal swabs isolated *Klebsiella*, which was resistant to all antibiotics tested except Amikacin, Mecillinam, and Meropenem. Tests for HIV, hepatitis B and C were all negative. After over a month of hospitalisation, her wound culture was positive for *Staphylococcus aureus*.

She was cared for in isolation under strict barrier nursing due to infection control concerns. Management was primarily conservative with analgesia, anticonvulsants, and NG feeding. IV antibiotics (Amikacin, Meropenem) were only given when she spiked temperatures and had raised inflammatory markers.

We continued to monitor her progress with input from the neuro-oncology, neuro-rehabilitation, and plastic surgery teams. Her GCS has improved and now remains at 15. Her biochemistry

parameters continued to oscillate throughout her admission. Prior to discharge, Hb was 103 g/L, platelets was $51 \times 10^9/L$, WBC was $2.4 \times 10^9/L$, urea was 8 mmol/L, creatinine was $41 \mu\text{mol/L}$. Final blood cultures remained negative. Her distal gangrene was initially managed conservatively with dressings but due to her prolonged admission in hospital for repatriation closer to home, it was decided for her to have a semi-elective focused debridement and amputation of selected digits of all four limbs. She has now recovered completely from this procedure and will be followed up by the plastic surgery team as an outpatient. She has been discharged with an ability to converse with her family and friends, obey commands in terms of motor response, and eat and drink a normal diet. She has been repatriated closer to her family and friends for on-going rehabilitation and therapy with a view of eventually being able to return to her own family home.



Fig. 3. Dorsal and plantar aspects of right foot.



Fig. 4. Dorsal and plantar aspects of left foot.

3. Discussion

While SPG, or purpura fulminans, is well documented in the literature, to our knowledge four limb SPG has not been previously reported as a postoperative complication following emergency neurosurgery. The phenomenon in this case was precipitated by characteristic triggers of SPG including bacterial septicaemia, DIC, and AKI—all of which are known complications of neurosurgery. It is therefore crucial to be mindful of SPG in the septic, postoperative neurosurgical patient. SPG should be suspected at the first sign of marked coldness, pallor, cyanosis or pain in the extremity, together

with increase in serum lactate levels. This condition can progress rapidly to frank gangrene without timely intervention.

The common organisms responsible for post-operative septicemia are *Pneumococcus*, *Staphylococcus*, *Streptococcus*, and Gram-negative organisms [6]. Although it is often challenging to isolate the cause of the vascular occlusion in SPG, it is speculated that the low-flow state, which is present in most cases due to low systemic vascular resistance, leads to thrombosis and occlusion of the microcirculation. In the early stages, pulses may still be palpable and the large vessels are often spared. In fact, pathological examination of amputated specimen has shown thrombi concentrated

in the small vessels with no thrombus in the large vessels. As distal extremities are especially susceptible, these ischemic changes begin distally and may progress proximally to involve the entire limb [2]. DIC is seen in up to 85% of cases of SPG, and clinical evidence is increasingly suggestive of DIC as the final common pathway of SPG pathogenesis [7].

No treatment has been found to be completely effective. Early recognition remains the key factors in SPG management. When peripheral perfusion is uncertain, aggressive fluid resuscitation is recommended with the aim to discontinue or reduce vasopressor therapy at the earliest possible chance. At the same time, treatment of the sepsis and DIC with IV antibiotics and low-dose heparin respectively (if feasible and applicable) should be promptly initiated [7]. Sympathetic blockade, IV vasodilators, local injection or IV infusion of alpha-blockers, and IV prostaglandins have all been attempted after appearance of digital ischemia to variable degree of success [4,5,8]. However, prevention is still the best line of management. In addition, caution is necessary when performing arterial punctures, as this may worsen the micro-thrombosis process [9].

An initial nonsurgical approach allows time for the patient's condition to stabilise and for the gangrene to become well demarcated. Amputation of the gangrenous tissue(s) may be inevitable, but is rarely required urgently [5]. Local debridement and secondary skin grafting are other treatment options to facilitate regaining of function and rehabilitation.

4. Conclusion

Postoperative septicaemia and secondary SPG are uncommon but important complications of neurosurgery. Awareness, early recognition, and prompt management are essential to avoid this condition. Routine examination of and vigilance of early signs of distal ischaemia is recommended. If SPG is diagnosed, an initial non-surgical approach is recommended, focusing on correcting the underlying cause(s). Long-term management is primarily supportive and symptomatic, and requires coordination via skilled

multidisciplinary teams following treatment of the acute problem(s), before surgical intervention is attempted. Appropriate postoperative care, primary management, and rehabilitation can lead to very successful outcomes.

Consent

Written informed consent was obtained from the patients' next of kin for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Conflict of interest

The authors report no conflicts of interest in this work.

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