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Case Report on Super-Refractory Status Epilepticus

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ABSTRACT

Introduction

Super-refractory status epilepticus (SRSE) is a critical condition that poses a serious threat to a patient's neurological outcome, with a high mortality rate, and is defined as a status epilepticus that persists despite 24 hours of treatment with an IV anesthetic, with the possibility of recurrence once the patient is weaned off the anesthetic medication. In Cambodia, there are no documented case reports of SRSE, nor are there established protocols to guide physicians, particularly emergency doctors and intensivists, in managing this condition. This case report aims to raise awareness, highlights diagnostic challenges, and discusses appropriate management strategies in resource-limited settings.

Case Presentation

A 21-year-old Cambodian male patient was admitted to the Medical Intensive Care Unit (ICU-Med) at Khmer-Soviet Hospital because of prolonged unmanageable seizures for four days following fever and extreme headache, with a visual analog score (VAS) of 9/10. The patient had been known to have had epilepsy since his first year of life. Owing to persistent seizures and unresponsiveness to the treatment, the patient was then transferred to the ICU-Med for the management of refractory status epilepticus.

Conclusion

SRSE is extremely rare, and its diagnosis and management are challenging. This case report provides the first evidence to spread the message, findings, management approach, and patient outcomes. The message is that even in a resource-limited situation, we are still able to manage the condition effectively.

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Introduction

Status epilepticus (SE) is characterized by a prolonged seizure episode lasting more than 30 minutes because of a failure of the brain's seizure-regulating mechanisms [1]. Such neurological abnormalities can lead to long-term complications, from neuronal death to neuronal injury, and affect the neuronal network, depending on the type of seizure and duration [1]. Even though SE can pose a serious threat to neurological function, its management follows a structured approach that includes assessing the airway, breathing, and circulation, as well as administering antiepileptic drugs (AED), with the main purpose of terminating the seizure while supporting the patient's cardiovascular and respiratory stability [2,3]. When a patient does not respond to one of the first-line medications (IV benzodiazepine) or one (IV AED), refractory status epilepticus (RSE) occurs [4]. However, super- refractory status epilepticus (SRSE) is a more critical condition that poses a serious threat to the patient's neurological outcome, with a high mortality rate, and is defined as an SE that persists despite 24 hours of treatment with an IV anesthetic, with the possibility of recurrence once the patient is weaning off the anesthetic medication [5,6]. In Cambodia, there are no documented case reports of SRSE, nor are there established protocols to guide physicians, particularly emergency doctors and intensivists, in managing this condition. This case report aims to raise awareness, highlight diagnostic challenges, and discuss appropriate management strategies in resource-limited settings.

Case Presentation

A 21-year-old Cambodian male patient was admitted to the Medical Intensive Care Unit (ICU-Med) at Khmer-Soviet Hospital because of prolonged unmanageable seizures for four days following fever and extreme headache, with a visual analog score (VAS) of 9/10. The patient had been known to have had epilepsy since his first year of life. He was initially treated at the Internal Medicine Department with Levetiracetam and Valproate, yet the seizure remained. Diazepam (20 mg) was subsequently administered at 30-minute intervals. He was then transferred to the Neurology Department for further management, where phenytoin at a loading dosage of 900 mg was given and reduced to 300 mg/day the following days, along with levetiracetam 750 mg BID and valproate 500 mg every 6 hours for two days.

Owing to persistent seizures and unresponsiveness to the treatment, the patient was then transferred to the ICU-Med for the management of refractory status epilepticus. Upon examination, the patient was somnolent, but the Glasgow score (GCS) was 13/15, with continuous tonico–clonic seizures, fever, and extreme headache. He received further physical examination, which revealed a positive Babinski sign on the left side of the feet, yet negative Kernig and Brudzinski signs were observed. The intensivist decided to have him intubated under general anesthesia, and thiopental 2 mg/kg/h was administered. Unfortunately, the seizures persisted for an additional 3 minutes. In that condition, Curare (Norcuron) was given to have him stabilized.

The laboratory findings of complete blood count (CBC), inflammatory marker (CRP), procalcitonin, glycemia, liver function test, and renal function were normal. To further investigate this phenomenon, a brain CT was performed, and the results revealed normal findings. Lumbar puncture was performed twice within three days, and the results revealed no abnormalities in protein concentration (day 1: 0.45 g/L; day 3: 0.31 g/L), glucose concentration (day 1: 0.71 g/dL; day 3: 0.72 g/dL), or WBC count (day 1: 03; day 3: 00). Further investigation of antinuclear antigen (ANA) was performed to rule out other autoimmune diseases, and the results were within the normal range (30.98 AU/ml).

Given the clinical findings and normal laboratory findings, the patient was diagnosed with super-refractory status epilepticus due to an idiopathic cause. He was stabilized with 5 anti-epileptic drugs (phenytoin, valproate, levetiracetam, thiopental, and ketamine) and admitted to the ICU-Med under

mechanical ventilation for 27 days. The convulsion began to subside by the 10th day of admission at ICU-Med. However, prolonged mechanical ventilation led to diaphragm muscle paralysis, resulting in the patient's inability to breathe spontaneously. Tracheostomy was performed to facilitate continued mechanical ventilation, physiotherapy, and nutritional support. Kinesitherapy was attempted daily to improve generalized muscle strength and respiratory rehabilitation. The patient's condition began to improve noticeably by day 27, when he was able to breathe independently and experienced only occasional brief convulsions of less than 30 seconds per day. He was then switched to oral anticonvulsants, including phenytoin, valproate, levetiracetam, and lorazepam. Once clinical stability was achieved, he was then transferred back to the Neurology Department for continued management and monitoring. Notably, despite regaining consciousness, the patient continues to experience long-term neurological sequelae because of a history of recurrent seizures, including motor deficits and cognitive impairment.

Discussion

The management of SRSE is crucial because it is life-threatening and remains a major management challenge for physicians, especially in a resource-limited context. The mortality rate can range from 30% to 50% with comorbidities [6,7]. Numerous studies have shown that mainly healthy young adults with a history of repetitive refractory seizures tend to have idiopathic seizures [8–10]. Ideally, when the root cause cannot be identified, electroencephalogram (EEG) and magnetic resonance imaging (MRI) can assist in diagnosis and interpretation with caution in terms of prognosis and outcome [11]. A systematic review and meta-analysis of outcomes and treatment approaches for SRSE by Cornwall et al. revealed no difference between sexes among 266 studies in terms of disease vulnerability [12]. The majority of the causes were acute cerebral events (41.2%), and 22.3% were unknown causes, with a mean duration of SRSE of 36.3 days [12]. These findings were similar to those of our case report, in which our patient was admitted to the medical ICU for 27 days, and the reason behind the patient's SRSE condition was idiopathic. The limitation of this case report is that CT scans failed to detect the diffusion/perfusion area to indicate whether there was sufficient blood flow within the brain. These findings could be detected only through MRI, to which the ICU-Med department did not have access.

Conclusion

SRSE is extremely rare, and even the highest hierarchical research, such as systematic reviews and meta-analyses, still suffers from a low level of evidence. This case report provides the first evidence for the spread of information, findings, management approaches, and patient outcomes. The message is that even in a resource-limited situation, we are still able to manage the condition effectively. This report should provide evidence to increase awareness among all physicians and intensivists in the context of Cambodia.

Contribution

Dr. Channa Prum and Dr. Theanhtheoun Molika were responsible for the writing of this manuscript, and Dr. Chendavatey Pok assisted and edited this case report.

Declaration

There are no conflicts of interest. Informed consent was obtained from the patients.

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