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Case Report

Hemispherotomy for late post-traumatic super-refractory status epilepticus in an adult

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1. Introduction

Super-refractory status epilepticus (SRSE) is a neurological emergency with extremely high rates of mortality and morbidity. Although acute severe brain injury is often cited as an etiology of this rare entity, there is no report of late posttraumatic seizures presenting as SRSE to the best of our knowledge. There are no established treatment guidelines for SRSE and the surgical intervention is usually considered very late when medically induced coma fails. Here we share our experience of treating a case of late post-traumatic SRSE of hemispheric origin, refractory to medical measures which were finally controlled effectively by hemispherotomy.

2. Case report

A 56-year-old gentleman was brought to the emergency department with generalized convulsive status epilepticus (GCSE) of 2 h duration. His airway was maintained and vitals were stable with a GCS (Glasgow Coma Scale) score of 10/15 (E2 V3 M5). Pupils were 2 mm bilateral and reactive to light.

He had sustained a severe head injury (GCS score 5/15) following a road traffic accident one year prior. CT brain had revealed a right temporoparietal acute subdural hematoma and a right frontal hematoma with a significant midline shift to the left requiring right sided decompressive hemicraniectomy [Fig. 1A]. His postoperative stay was uneventful and he has discharged with a GCS of 14 and residual left hemiparesis. He was on regular follow-up in the brain injury clinic for a year and had a Glasgow outcome scale (GOS) of 5. He was compliant with medications including his anti-seizure drug, phenytoin.

Routine laboratory investigations like serum electrolytes, glucose, magnesium, calcium, liver function tests, complete blood cell count, creatinine and anti-seizure medication levels were sent and found to be normal.

In view of ongoing seizures, his airway was secured and the initially managed with phenytoin and levetiracetam at higher blood concentrations. Since there was no response, we started him on Midazolam, a short-acting benzodiazepine, at a loading dose of 0.2 mg/kg and maintained by a continuous infusion of 0.05 to 2.0 mg/kg per hour. CT brain was repeated, which showed no significant abnormality except for a craniectomy defect of previous surgery and slightly bulging brain through that defect. EEG revealed right frontal and parietal epileptiform discharges.

He was managed with midazolam infusion and boluses and maximal doses of anti-seizure drugs including phenytoin, levetiracetam, clobazam. He was also initiated on thiopentone infusion (barbiturate coma). The barbiturate coma was maintained for 48 h during which time he would remain seizure free and seizures recurred during tapering of thiopentone infusion. Thus further therapeutic agents were added including topiramate and lacosamide along with supplementation with folate, pyridoxine, thiamine and a ketogenic diet.

Further diagnostic evaluation with CSF and blood investigations was negative for infective and metabolic causes of seizures. Imaging with MRI brain showed gliotic areas in the right frontal and parietal lobes which denoted the sequel of previous trauma [Fig. 1B and C].

Electroencephalography (EEG) was repeated at the bedside in the Intensive Care Unit, by a portable Galileo machine, (EB Neuro S.p.A., BE Light, Firenze, Italy). Scalp recordings using 10–20 system of electrode placement showed theta range background with right breach rhythm and lateralized periodic discharges (LPDs), [Fig. 2A].

His sensorium fluctuated between a GCS score of 14 to 14 and he continued to have about 20–30 episodes of seizures every day (in the form of twitching involving the left upper limb and lower limb) even on barbiturate coma. Having failed to withdraw the barbiturate coma after one week, we decided to perform a right peri-insular hemispherotomy considering the underlying right hemispheric damage. The family members were explained in detail the patient's clinical condition and the procedure was planned and consent was obtained. The patient underwent a right peri-insular hemispherotomy. In the immediate postoperative period, his sensorium improved to a GCS score of 14 and he remained seizure free. Postoperative EEG showed the similar background with a breach rhythm from the right hemisphere [Fig. 2B]. On the third postoperative day, we were able to stop the midazolam infusion. In the immediate postoperative period, his sensorium improved

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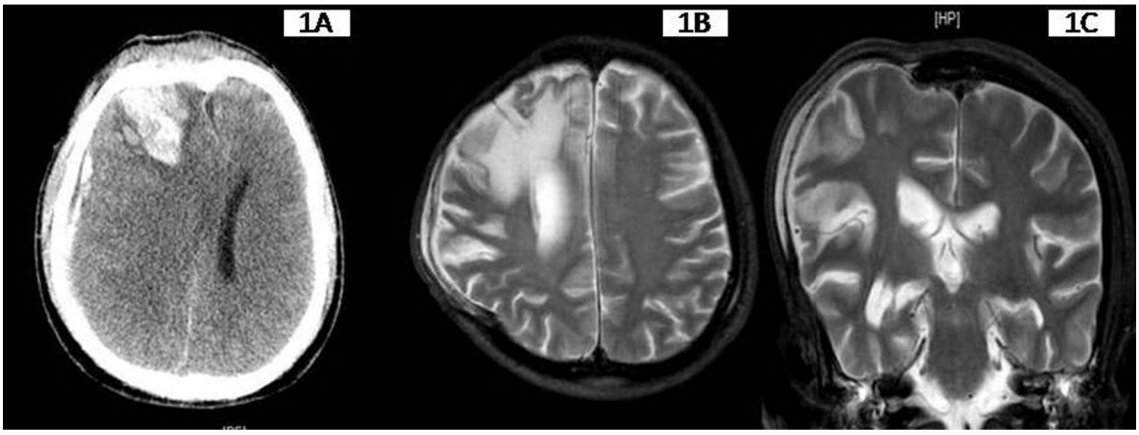

Fig. 1. A. Preoperative CT Scan brain showing right-sided temporoparietal acute subdural hematoma and a right frontal hematoma with a significant midline shift to the left. B. Postoperative magnetic resonance imaging (MRI) brain showing focal gliotic area with long TR weighted hyperintensity in the right frontoparietal region, related to post the traumatic sequel. C. Postoperative MRI brain showing asymmetry of hippocampus without any signal changes.

to a GCS score of 14. His GCS gradually improved to 14 on the fifth post-operative day. He was discharged on the thirteenth postoperative day with a GCS of 11. At 3 months follow up, his GCS was 14 and could walk with support. There were no further seizures.

3. Discussion

Seizures are well-recognized complications of the traumatic brain injury (TBI), which are divided into early, occurring within the first

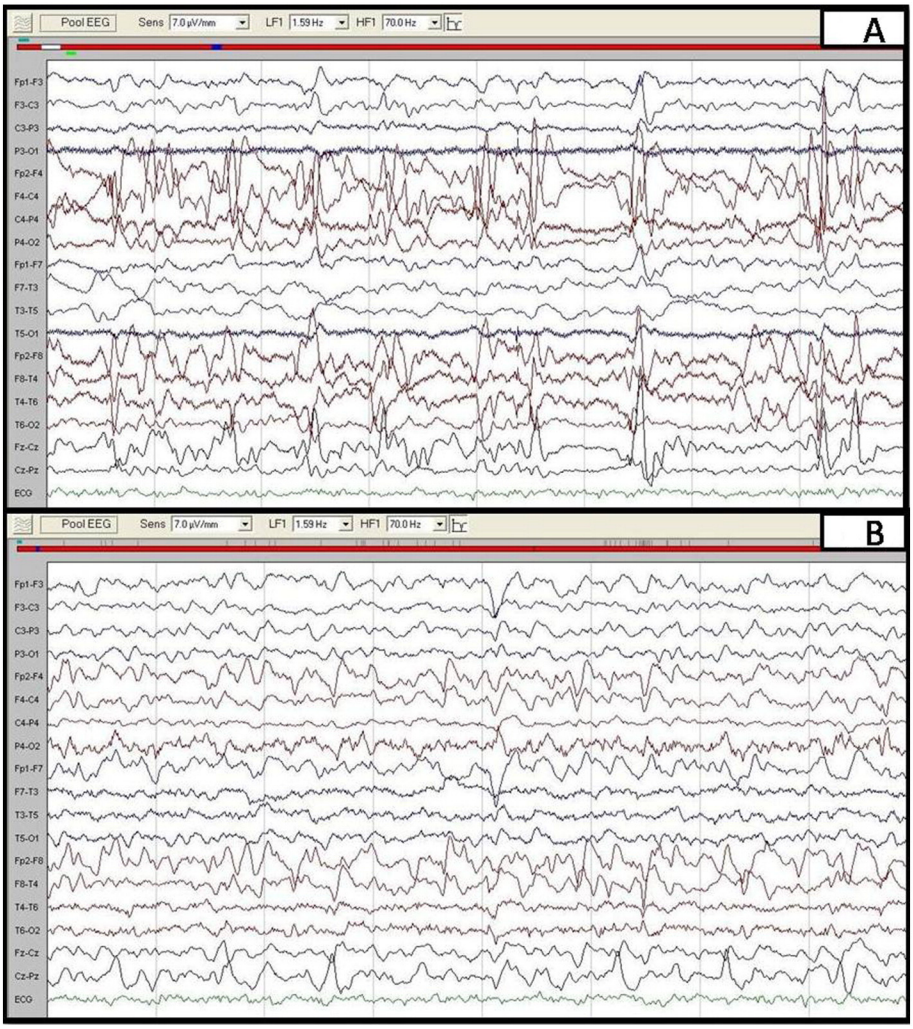

Fig. 2. A. Preoperative Electroencephalography (EEG) showing lateralized periodic discharges (LPDs) (arrows). B. Postoperative EEG showing no discharges under the breach rhythm in the right hemisphere. Filter settings were high pass at 1.59 Hz and low pass at 70 Hz, and sensitivity at 7 µ volts/mm.

week of injury and late posttraumatic epilepsy (LPTE), with onset 7 days post injury [1].

The estimated incidence of seizure in severe head injury is 10% [1] and around 1.9% to 8% of these patients develop status epilepticus (SE) [2]. The incidence of LPTE ranges from 5 to 19% [3–5]. The data on the incidence of refractory status epilepticus (RSE) is scarce and there is no report of late-onset post-traumatic super-refractory status epilepticus in the literature to the best of our knowledge. There are multiple factors associated with the occurrence of LPTS such as age, the severity of an injury, the location of an injury, penetrating injuries, the presence of hematoma, the presence of metal fragment and occurrence of an early traumatic seizure [3–5]. The estimated risk of developing epilepsy within the first year of penetrating head trauma is found to be 580 times higher compared to a population not at risk [6].

Status epilepticus (SE) is a medical emergency. Estimated Incidence of SE in Europe and US is around 20/100,000 per year [7]. SE is considered refractory when there is no response to two to three anti-seizure drugs. The mortality rate is three times more in RSE compared to SE [8]. SRSE is defined as status epilepticus that continues or recurs 24 h or more after the initiation of anesthetic therapy, including those cases where status epilepticus recurs on the reduction or withdrawal of anesthesia [9]. About 15% of all status epilepticus progress to super-refractory status epilepticus [9]. Neurological outcome of the patients on long-term medically induced coma is very poor. Prolonged suppressive therapy to control the seizure has got deleterious effects on the nervous system [10]. Recent reports suggest the role of neurosurgery in controlling some super-refractory status epilepticus either in the form of hemispherectomy, hemispherotomy or its modifications [11].

The first hemispherectomy for epilepsy was performed by McKenzie [12] in late 1930's. In 1950, the first series of anatomic hemispherectomy in epilepsy was performed for infantile hemiplegia mainly in the pediatric population [13]. To minimize the invasiveness of the functional hemispherectomy, multiple variations in the disconnections have been proposed [14]. Most popular type is called trans-Sylvian keyhole functional hemispherectomy described by Schramm [15].

Hemispherectomy for the refractory status epileptics was first reported in 2004 where anatomic hemispherectomy was done on a 7-year boy with cortical dysplasia in RSE [16]. The first 2 adult cases of hemispherotomy for RSE was reported by Oderiz et al. in 2015 [17] followed by two more adult cases reported by McGinity M et al. in 2016 [11]. Until now, only four cases of hemispherotomy for RSE in adults have been reported [11].

Our case is unique because of two reasons. First, the hemispherotomy for RSE was performed in an elderly patient of 68 years (to date only four adult cases have been reported). The second reason is because this case is the first example of late-onset post-traumatic super-refractory status epilepticus. Our decision of surgical intervention was based on the failure of response to the barbiturate coma even after 10 days and the complications associated with the long term therapeutic coma (ventilator-associated pneumonia, deep vein thrombosis and ileus with micro perforations). Thus our patient underwent a right sided non-dominant hemispherotomy and had a very good neurological outcome following the surgery.

Although the hemispherotomy was a major surgical procedure, in view of complications associated with the long-term therapeutic coma and lack of other options, we went ahead with the procedure. Although the outcome was very good in this patient, it would be too early to generalize from a single case experience.

4. Conclusions

SRSE, although rare as the primary post-traumatic disability, can occur in post-traumatic head injury patients outside of the acute period of injury. Hemispherotomy may be considered as an effective therapeutic option in patients with SRSE who fail to respond to medical management. Further studies are required to assess how early this option should be considered in the management of the adult patients with late post traumatic SRSE.

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