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RESEARCH ARTICLE

The Syndrome of Trephined: Role of the Anesthesiologists in Overcoming the Physiological Changes

Widiartha Wahyudi[1], Dhania A. Santosa[2], Bambang Pujo Semedi[3], Christrijogo Sumartono Waloejo[4*], Kohar Hari Santoso[5]

[1, 2,3,4,5] Department of Anesthesiology and Reanimation, Universitas Airlangga, Dr. Soetomo General Academic Hospital, Surabaya, Indonesia

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***Corresponding Author:**

christrijogo@fk.unair.ac.id

ABSTRACT

A 58-year-old male with a skull defect came to the Emergency Department with a history decompressive craniectomy caused by subdural hematoma. 5 months after the surgery he came back with general weakness. His consciousness progressively decreased to GCS 4 (E2VxM2). MRI showed brain edema and midline shift to the right side suggesting a Syndrome of the Trephined. He underwent autograft cranioplasty to close the defect. After the surgery, the patient's general condition improved significantly. The consciousness and limb weakness began to improve, he was discharged after 3 days. The clinical spectrum of Syndrome of the Trephined shows symptoms of increased intracranial pressure such as headache, decreased consciousness, and other neurological deficits. Anesthesiologists hold a vital role in managing the physiology changes, deciding the most suitable anesthesia technique, and choosing the right medication to overcome this physiological changes. Management in these patients focused on reducing the compressive effects of atmospheric pressure on brain tissue. Cranioplasty is the definitive therapy for Syndrome of the Trephined.

INTRODUCTION

Syndrome of the Trephined is often associated with complications of craniectomy. Syndrome of the Trephined was first described by Grant in 1939 as a collection of symptoms that included seizures, dizziness, headache, mental depression or discomfort at the defect site, which would resolve after cranioplasty surgery. Yamaura and Makino used the term "Sinking Skin Flap Syndrome" to describe the same clinical cluster, in patients with skull bone defects after craniectomy. Yamamura et al. defined this condition as a neurological disorder caused by the direct compressive impact of atmospheric pressure on the brain tissue underneath the skull defect. Several studies have subsequently reported several of clinical presentations of the syndrome, with motor weakness being the most common symptom, followed by cognitive impairment and language impairment, impaired alertness, headaches, seizures, cranial nerve defects, vertigo, and mood disorders.

This case was found in November 2022 at Dr. Soetomo General Academic Hospital Surabaya, Indonesia. A 58-year-old man presented with a chief complaint of generalised weakness, with a history of four surgeries, and an inwardly concave left skull defect. During hospitalization the patient's consciousness progressively decreased and diagnostic impressed the complication of Syndrome of the Trephined. The patient underwent cranioplasty surgery to reduce the effects of atmospheric pressure to the brain tissue and the patient's condition improved significantly after surgery.

MATERIAL AND METHODS

About 7 months earlier, the patient complained of sudden loss of consciousness accompanied by vomiting and seizures. A contrast-enhanced CT scan showed an intraventricular haemorrhage (IVH) and hydrocephalus. Then the patient underwent surgery to insert an External Ventricular Drain (EVD). 1 week later, the patient underwent a second surgery to change the EVD system to VP shunt. The patient was then discharged and was able to ambulate.

One month later the patient came to the hospital with a chief complaint of decreased consciousness 7 hours before admission. Initially, the patient complained of a headache accompanied by vomiting 3 times. The patient's consciousness decreased gradually and became increasingly severe, making it difficult to speak and open his eyes. CT scan diagnostic results showed a subdural hemorrhage in the fronto-temporo-parietal region sinistra, midline shift as far as 1.3 cm towards the left side, and slit ventricle. Then the patient underwent craniotomy to evacuate subdural haemorrhage, osteoplasty, and shunt ligation due to suspicion of cerebrospinal fluid overdrainage. Since then the patient is on total bed rest.

The patient's consciousness worsened 2 days after surgery with a decrease in GCS of 5 (E1V1M3) and anisocor pupils. CT scan evaluation showed subdural haemorrhage in the fronto-temporo-parietal region sinistra which suggested re-bleeding from the previous surgery. The patient underwent re-open craniotomy surgery to evacuate the subdural haemorrhage and decompressive craniectomy. After surgery the patient's consciousness improved and was discharged to follow a rehabilitation programme.

Five months later, the patient came back to the hospital with a left skull defect from the previous surgery (Figure 1). The patient was conscious with GCS E4VxM5 related to aphasia. There was weakness in the right extremity. During hospitalization, the patient's consciousness decreased to GCS E2VxM2.

Figure 1. Clinical Presentation

Physical examination showed no abnormality with the airway, no respiratory and hemodynamic problems, GCS was 2x2 related to aphasia, no urinary and digestive problems and the patient's temperature was within normal limits.

Blood laboratory examination results showed hypoalbumin 2.39 g/dL. ECG examination and chest X-ray are within normal results. The results of AP and lateral skull photos showed an osteotomy defect of the calvaria in the left fronto-temporo-parietal region with bone osteotomy fragments projected in the left parietal region, and a medical device (VP shunt) with proximal tips projected intracranially. Head MRI revealed a bony defect in the left fronto-temporo-parietal region, perifocal oedema in the left centrum semiovale and left corona radiata squeezing the left lateral ventricle and a 1.5 cm midline shift to the right hemisphere (Figure 2).

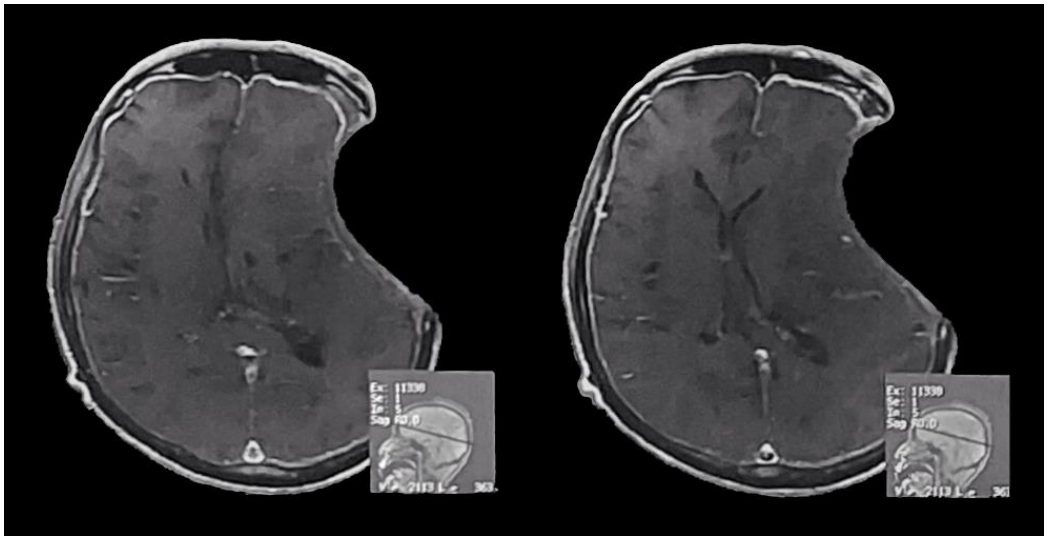

Figure 2. Head MRI showed bone defect in the left fronto-temporo-parietal region, perifocal oedema in the left centrum semiovale and left corona radiata and 1.5 cm midline shift to the right hemisphere

The patient underwent autograft cranioplasty surgery under general anesthesia with intubation airway management, combined with scalp block. Preoperative vital signs were within normal range. Preoxygenation with 100% Oxygen was carried out to maximize oxygen reserve. Lidocaine was administered 3 minutes before induction to minimize sympathomimetic response to laryngoscopy and endotracheal intubation. Fentanyl was given before induction as an opioid-based analgesic agent. One minute after Fentanyl administration, Propofol was given to induce hypnosis until loss of consciousness was confirmed. Then, manual control of ventilation was carried out. Rocuronium was given as a muscle relaxant after ventilation was confirmed adequate. After that, smooth tracheal intubation followed by scalp block with Ropivacaine 0.5% were performed to minimize opioid use. Volatile agent Sevoflurane were used to maintain the anesthesia intraoperatively. The surgery lasted for 4 hours, hemodynamics were stable with bleeding of 150 ml, and there were no adverse events during the surgery. The patient was then extubated at the end of the operation.

After surgery the patient was admitted to the intensive care unit for close monitoring. The diagnostic results of the postoperative evaluation suggested a decrease in midline shift. The consciousness started to improve, pupils were isochore and limb weakness started to decrease. The patient's condition gradually improved significantly and the patient was discharged to continue rehabilitation.

DISCUSSION

There are several pathophysiologic theories regarding the cause of symptoms in Syndrome of the Trephined (Ho et al., 2020), some of which are: (a) Compression of the Cortex Due to Scalp Defect: Compression of the cortex in the defect area by the scalp is the cause of the symptoms that occur in this syndrome; (b) Cerebrospinal Fluid Flow; and (c) Impaired Brain Metabolism.

The aim of treating patients with this syndrome is to correct the effects ofatmospheric pressure caused by direct pressure of the area where the previous craniectomy procedure was performed. With the alleviation of this pressure effect, neurological symptoms will gradually improve. This can be achieved in several ways, conservative therapy including intravenous fluids, avoidance of hyperosmotic fluids, head down position (Trendelenburg position) or head turn to the side of the craniectomy can gradually reduce neurological deficits. Therapy involving surgery can include shunt ligation if there is still a shunt device and when there is a suspicion of shunt overdrainage. The definitive treatment for Syndrome of the Trephined is cranioplasty, which has been shown to improve neurological deficits and correct abnormal cerebrospinal fluid flow. Cranioplasty can improve postural blood flow regulation, cerebrovascular reserve capacity, and cerebral glucose metabolism, with a positive impact of improved perfusion and hemodynamics. There is still no literature that determines the best time to perform cranioplasty but a study showed that there is no correlation between clinical neurology improvement and the interval of cranioplasty from a previous craniotomy procedure. The study by Nancy et al. stated that 94.11% of patients experienced clinical improvement after undergoing cranioplasty. The mean time for improvement of these neurological deficits was 6.5 days (Santander et al., 2022).

Min et al. reported a case of Syndrome of the Trephined caused by overdrainage of the lumbar-peritoneal shunt. The patient had semicomatose deterioration of consciousness, dilated pupils and decreased light reflexes. Shunt ligation was performed to prevent further drainage but the improvement was not significant. The patient then underwent cranioplasty surgery, and on the following day the light reflexes returned and the patient's consciousness significantly improved.

General anesthesia with endotracheal intubation airway management was used in the anesthetic management of this patient. Propofol were used at induction as a sedative agent. Propofol provide smooth induction and has been shown to possess neuroprotective properties, potentially reducing neuronal injury and improving outcomes in neurosurgical patients. An alternative drug is Thiopental which is a Barbiturate, which can reduce Cerebral Blood Flow (CBF) and Cerebral Oxygen Metabolic Rate (CMRO2) according to the dose given. The effect of barbiturates on reducing CBF and CMRO2 will cause a decrease in ICP (Halimi et al., 2019).

Scalp block was performed for analgesia during and after the surgery. Based on recent study, scalp block using 0.5% Ropivacaine obtain preferable postoperative analgesia compared to lower concentrations. Scalp block with Ropivacaine also reduced hemodynamic fluctuations in craniotomy operations (Yang et al., 2020). General anesthesia combined scalp block with ropivacaine 0.5% in craniotomy patient is more effective in reducing MAP and heart rate during scalp incision and periosteal contact compared with general anesthesia alone (Iskandar Suryadani et al., 2020). Utilization of the scalp block also appears to improve pain control and lessen opioid use in the 4 hours of the immediate post-operative period (Patel et al., 2021). Another study also conclude that scalp block using Ropivacaine 0.5% preincision is more effective in reducing pain scale up to 12 hours and also reduced the requirement of Fentanyl within 24-hours post craniotomy compared to general anesthesia alone (Muhammad Aviv Pasa et al., 2020). Although scalp block is contraindicated in patient with absence of bone flap due to previoius craniotomy, this procedure is still performed but with intense caution and no injections were done around the area of the scalp defect.

Intraoperatively, Sevoflurane was chosen instead of Total Intravenous Anesthesia (TIVA). Propofol can cause significant hypotension, which may not be desirable in patients with compromised cardiovascular function. Sevoflurane generally has less impact on cardiovascular function compared to Propofol, making it a preferred choice in patients with compromised cardiac function. Sevoflurane also has an adjustable depth of anesthesia, it allows for easy titration of anesthesia depth by adjusting the concentration of the vaporizer, providing flexibility during surgery. In neuroanesthesia, Sevoflurane is beneficial in preserving cerebral autoregulation. Postoperatively, rapid offset of Sevoflurane should facilitate early evaluation in the neurosurgical setting. The cost-effective analysis also showed that Sevoflurane was less expensive than propofol (Rehi, 2023).

Nitrous Oxide in the other hand, has a stirnulatory effect on the brain and increases Cerebral Blood Flow (CBF) in excess of its effect on metabolism. During the anesthetic administration, a significant amount of N2O enters closed gas spaces. These spaces mainly contain nitrogen (from air), which has low blood/gas partition coefficient (0.013) that limits its removal by blood. Thus, the rate of entrance of N2O into the closed gas spaces is significantly more than the exit of nitrogen from the closed gas spaced to the blood stream, resulting in a rapid increase in volume. In a nondistensible cavity like cranial vault, such rapid increase of volume could result in "tension pneumocephalus" especially with preexisting pneumocephalus. Based on this argument, it is recommended to avoid N2O altogether or discontinue its use prior to the dural closure.

In contrast, awake cranioplasty is also widely used. It involves keeping the patient awake and responsive during the surgery. Scalp block in awake craniotomy makes the patient tolerable for the surgery and enables better patient satisfaction after surgery in terms of pain compared with general anesthesia. Intraoperatively, patient can be sedated using intravenous Dexmedetomidine. Dexmedetomidine preserves the respiratory drive, maintains heart rate, provides analgesia, decreases cerebral metabolic rate of oxygen, and prevents CO2 retention, thereby maintaining cerebral blood flow. Dexmedetomidine also decreases opioid dose and reduces new-onset postoperative AF.

Hemodynamic management and monitoring was achieved by maintaining blood pressure and pulse within 20% of baseline. Ventilation and fluid management were also key considerations, in this case tidal volume was maintained at 6 to 8 mL/kg and PEEP was set to 5 cmH2O to maintain oxygenation and ventilation. Excessive PEEP should be avoided, because it can cause an increase in intrathoracic pressure, causing changes in cerebral venous drainage and an increase in ICP (Harijono et al., n.d.). The patient was kept in a state of euvolemia, overhydration should be avoided as it would lead to increased blood flow to the brain and aggravate the edema that already occurred. NaCl 0.9% was chosen for fluids due to its isotonicity and avoiding cerebri edema.

The emergence and extubation phases have special considerations in these patients. Extubation must be done carefully to prevent activation of the sympathetic nervous system which can lead to increased intracranial pressure. Administration of Propofol, Remifentanil or Lidocaine may help prevent coughing and agitation during this process.

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DECLARATION OF INTEREST

The authors report no conflicts of interest in this work.

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