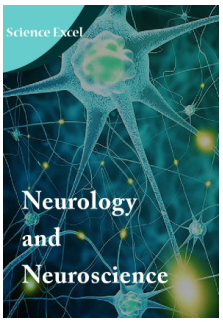
Neurology & Neuroscience

Case Report



Cranioplasty For Treatment of Syndrome of The Trephined: Case Report And Literature Review

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Abstract

Syndrome of the Trephined (ST) is a neurological complication observed after decompressive craniectomies, characterized by progressive neurological deterioration linked to the removal of a significant portion of the skull. ST commonly occurs in patients with severe traumatic brain injury, stroke, or other conditions causing intracranial hypertension. While rare, its clinical relevance is significant due to the impact on patient recovery. The syndrome's incidence varies depending on the clinical context, with risk factors including hemorrhagic injuries and brain displacement. In Brazil, its prevalence is underreported, partly due to the lack of large studies, though the incidence has risen with the increasing use of decompressive craniectomies in cases of severe intracranial hypertension. We present a case of a 69-year-old male patient with ST after a decompressive craniectomy for acute subdural hematoma and cerebral edema following trauma. Cranioplasty was performed to address the bone defect, and post-surgical recovery showed significant motor improvement, although swallowing difficulties persisted. The primary therapeutic approach for ST is cranioplasty, with early intervention shown to improve neurological outcomes. Early cranioplasty, performed within 30 days of decompressive craniectomy, leads to a recovery rate of up to 75%. This case highlights the importance of timely surgical intervention and the need for further studies on ST, particularly focusing on its epidemiology and pathophysiology in Brazil.

Introduction

Syndrome of the Trephined (ST) is a neurological complication observed after decompressive craniectomies, characterized by progressive neurological deterioration associated with the removal of a significant portion of the skull. ST is commonly seen in patients with severe traumatic brain injury, stroke, or other conditions that cause intracranial hypertension. Although rare, its clinical relevance is significant due to its impact on patient recovery. The incidence of ST varies depending on the clinical context and region, with risk factors such as hemorrhagic injuries and brain displacement. In Brazil, the prevalence of the syndrome is underreported, partly due to the lack of large-scale studies, but its incidence has been increasing as decompressive craniectomies are more commonly performed in patients with severe intracranial hypertension. In the United States, it is estimated that between 1% and 40% of patients undergoing decompressive craniectomies may develop ST, with a higher prevalence in cases of severe traumatic brain injury and stroke [1,2].

Case Report

A 69-year-old male patient with a history of being found unconscious after a horse kick underwent imaging tests at another hospital where an acute subdural hematoma and cerebral edema were detected, necessitating urgent decompressive craniectomy. A control CT scan was performed 45 days after the event (Figures 1 and 2), showing right-sided hemiparesis, a Glasgow coma scale (GCS) of 9 points, dysarthria, and swallowing difficulties.

Due to the clinical signs and symptoms, along with the CT findings, the patient was diagnosed with ST and surgical intervention for bone defect correction (cranioplasty) was indicated. The approach was made in the left frontotemporoparietal region, over the previous scar, with exposure of the bone margins and dura mater. (Figure 3)

A manual mold with orthopedic cement was created and fixed with mini plates and mini screws. (Figure 4)

Postoperative cranial CT scan with 3D reconstruction showing correction of the bone defect. (Figure 5)

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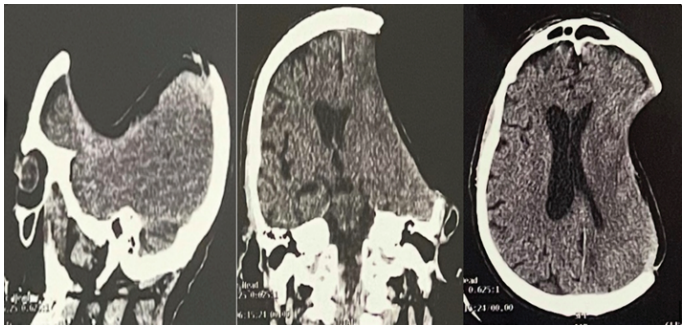

***Figure 1**. Sagittal, coronal, and axial cranial CT scan showing a bone defect due to decompressive craniectomy, with a midline shift and compression of the brain parenchyma.*

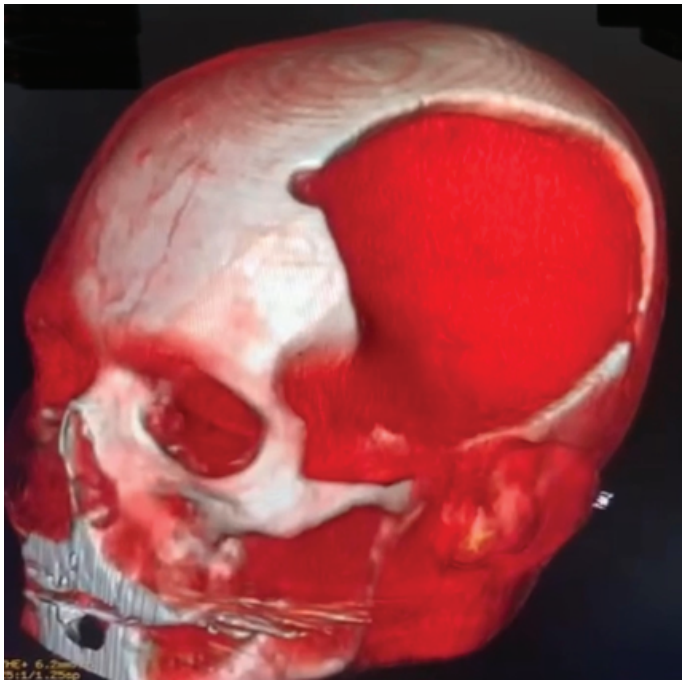

***Figure 2**. 3D reconstruction of the CT scan showing the craniectomy and depression of the brain parenchyma.*

One week after the surgery, the patient showed significant improvement in right-sided motor deficits, improved speech, and a GCS of 12 points, although swallowing difficulties persisted, requiring gastrostomy for feeding and prevention of aspiration. The patient is undergoing outpatient follow-up and is in the rehabilitation phase with motor physiotherapy.

Discussion

ST presents a range of clinical manifestations, including progressive neurological deterioration, motor, cognitive, and speech deficits. Additionally, patients often show temporary symptom improvement when changing to a supine position, a phenomenon known as orthostatic. While this suggests a possible relationship between the patient's position and intracranial dynamics, the exact mechanism behind this temporary improvement remains unclear [3].

The epidemiology ST varies by region and patient type. In the United States, it is estimated that the syndrome affects between 1% and 40% of patients undergoing decompressive craniectomies, with a higher prevalence in cases of traumatic

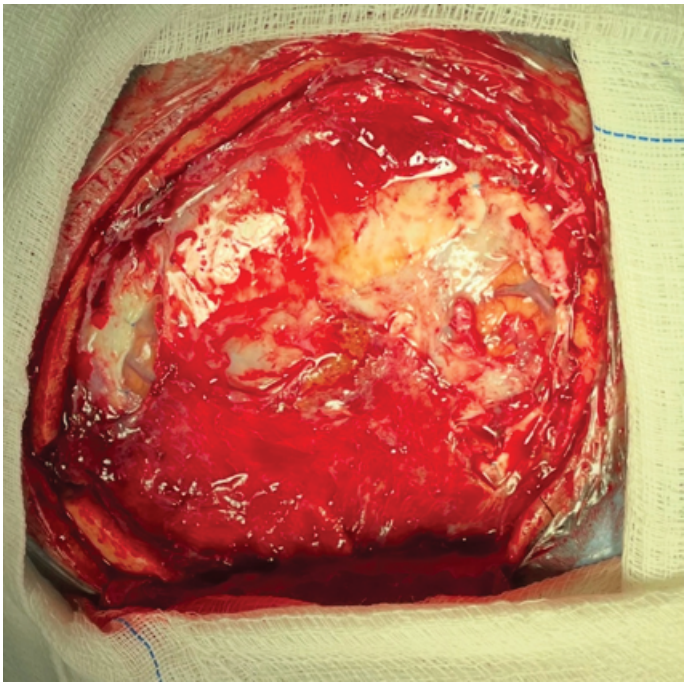

***Figure 3.** Intraoperative image showing the exposure of the bone margins and the dura mater covering the brain parenchyma.*

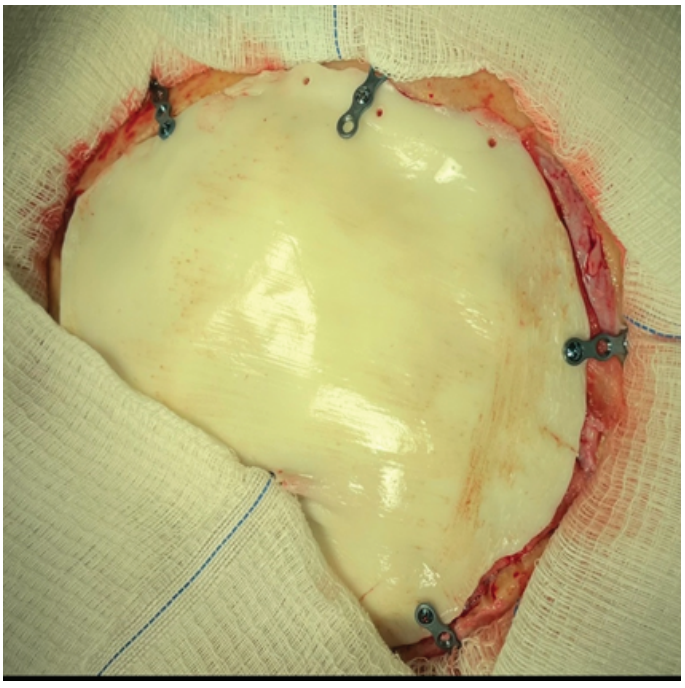

***Figure 4.** Intraoperative image showing the cranioplasty with bone cement and fixation with 4 mini plates and 8 mini screws.*

brain injury and stroke. A study by Wang et al. observed that approximately 25% of patients undergoing decompressive craniectomies for severe traumatic brain injury showed some degree of neurological deterioration related to the syndrome [4].

In Brazil, the condition is less discussed in terms of absolute numbers, but some local studies have observed an increasing prevalence, reflecting the rise in decompressive craniectomies for severe intracranial hypertension, especially in trauma

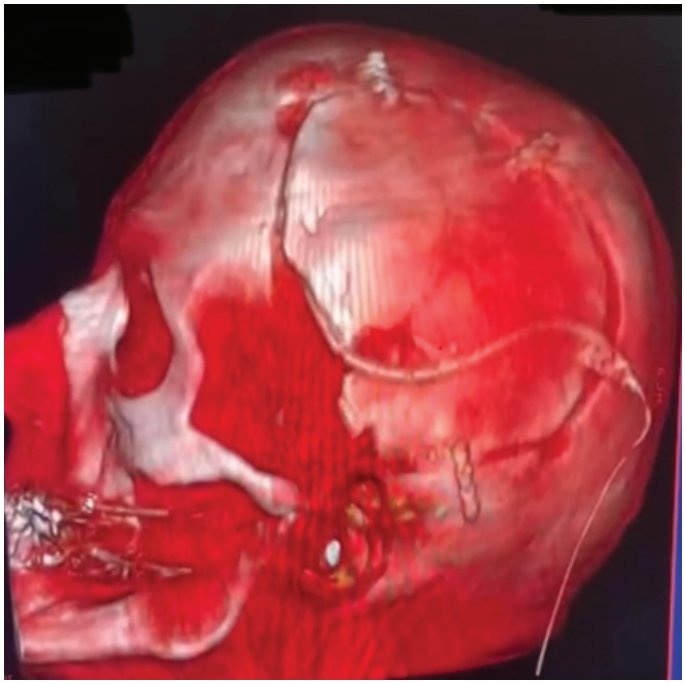

Figure 5. *3D reconstruction of the postoperative CT scan showing correction of the bone defect.*

patients [5]. The lack of systematic large-scale data collection in the country hampers a more accurate epidemiological analysis. Additionally, literature reveals that despite decompressive craniectomy being effective in reducing intracranial pressure, the risk of complications like ST increases as the amount of skull removed increases. In cases of bilateral craniectomy, the risk of developing the syndrome may be higher, as the absence of complete bone coverage facilitates the displacement of brain structures, potentially causing local negative pressure [6,7].

The primary therapeutic approach for ST is cranioplasty, aimed at restoring skull integrity and alleviating neurological symptoms. Early cranioplasty is a critical factor for clinical improvement. A multicenter study by Vaziri et al. showed that early cranioplasty, performed within 30 days of decompressive craniectomy, had a recovery rate of 75%, whereas patients waiting more than 90 days for the procedure had a recovery rate of only 40% [8].

Early cranioplasty not only helps restore neurological function but also reduces the incidence of additional complications, such as infections or new episodes of intracranial hypertension [9]. Studies indicate that, after cranioplasty, more than 90% of patients experience considerable improvement in cognitive and motor function, with favorable outcomes continuing to improve over time [10].

It is important to note that the syndrome may be more prevalent in elderly individuals, with a higher incidence of persistent neurological deficits following craniectomy. This may be related to increased brain fragility and reduced ability to adapt to the absence of a portion of the skull [11]. Furthermore, clinical response to treatment may be influenced by the severity of the original brain injury, with patients who have extensive brain damage showing slower or incomplete recovery [12].

Conclusion

ST is a relevant complication in the context of decompressive craniectomies and can have a significant impact on neurological recovery and patients' quality of life. Early diagnosis and appropriate therapeutic intervention, such as early cranioplasty, are crucial for clinical improvement. With proper treatment, most patients experience functional recovery, with improvement rates reaching up to 75% in cases of early cranioplasty. While understanding of the syndrome has evolved, there is still a need for more longitudinal studies, particularly focusing on epidemiology in Brazil and the pathophysiological mechanisms behind the observed orthostatic improvement and neurological deterioration in ST.

Conflict of Interests

The authors have no conflict of interests to declare

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