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Name of Journal: *World Journal of Clinical Cases*

Manuscript NO: 71733

Manuscript Type: CASE REPORT

Paradoxical herniation after decompressive craniectomy provoked by mannitol: A case report

Du C *et al.* Paradoxical herniation provoked by mannitol

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Abstract

BACKGROUND

Paradoxical transtentorial herniation is a rare but life-threatening complication of cerebrospinal fluid drainage in patients with large decompressive craniectomy. However, paradoxical transtentorial herniation after rapid intravenous infusion of mannitol has not been reported yet.

CASE SUMMARY

A 48-year-old male suffered from a right temporal vascular malformation with hemorrhage. In a coma, the patient was given emergency vascular malformation resection, hematoma removal, and the right decompressive craniectomy. The patient woke up on the first day after the operation and was given 50 g of 20% mannitol intravenously every 8 h without cerebrospinal fluid drainage. On the morning of the 7th postoperative day, after 50 g of 20% mannitol infusion in the Fowler's position, the neurological function of the patient continued to deteriorate, and the right pupils dilated to 4 mm and the left to 2 mm. Additionally, CT revealed an increasing midline shift and transtentorial herniation. The patient was placed in a supine position and

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given 0.9% saline intravenously. A few hours later, the patient was fully awake with purposeful movements on his right side and normal communication.

CONCLUSION

Paradoxical herniation may occur, although rarely, after infusing high-dose mannitol intravenously in the Fowler's position in the case of a large craniectomy defect. An attempt should be made to place the patient in the supine position because this simple maneuver may be life-saving. Do not use high-dose mannitol when the flap is severely sunken.

Key Words: Decompressive craniectomy; Intracranial hypotension; Paradoxical herniation; Transtentorial herniation; Mannitol; Case report

Du C, Tang H, Fan S. Paradoxical herniation after decompressive craniectomy provoked by mannitol: A case report. *World J Clin Cases* 2021; In press

Core Tip: The paradoxical herniation is a rare but life-threatening complication in patients with large decompressive craniectomies. This case report suggests that mannitol treatment after a large decompressive craniectomy can cause a paradoxical herniation. Early recognition and proper treatment can save a patient's life.

INTRODUCTION

The paradoxical herniation is a rare but life-threatening complication of cerebrospinal fluid (CSF) drainage in patients with large decompressive craniectomies. This result is due to a combined effect of brain gravity, atmospheric pressure, and intracranial hypotension^[1,2]. Paradoxical herniation has been reported after craniotomy in the context of CSF hypovolemia, usually ascribed to intraoperative or postoperative CSF drainage or spinal CSF fistula^[1,3-5]. Currently, only a few cases have been reported that paradoxical herniation may occur in the absence of CSF drainage, which is somewhat

different from our case^[6-8]. We describe a unique case of spontaneous paradoxical herniation after intravenous infusion of mannitol and decompressive craniectomy.

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CASE PRESENTATION

Chief complaints

A 48-year-old male was admitted to our hospital with sudden weakness in his left limb.

History of present illness

Two hours before his arrival, he suddenly developed weakness in his left limb and was unable to stand while working. The neurological examination showed a Glasgow Coma Scale score of 8 and a left hemiparesis. An urgent head CT examination revealed massive right temporal hemorrhage (Figure 1A and B). The patient underwent an emergent frontoparietal decompressive craniectomy with a duraplasty, and the hematoma was completely evacuated. A mass of abnormal blood vessels founded in the hematoma cavity during the operation was removed. Postoperative pathological examination revealed malformed blood vessels (Figure 1E and F). On the first day after the operation, the patient was fully awake with normal communication and directional movement of the right limb but had left hemiplegia. The head CT shows that the hematoma has been completely cleared, and the midline is almost in the middle (Figure 1C and D). The CT angiography shows normal cerebral vessels (Figure 1G and H). Although the flap pressure was not high, 50 g of 20% mannitol was given every 8 h to reduce local edema. On the morning of the 7th day after the operation, after an intravenous drip of 50 g of mannitol in the Fowler's position, the neurological function of the patient continued to deteriorate, the right pupil dilated to 4 mm and the left to 2 mm. An urgent head CT revealed increasing midline shift, transtentorial herniation, and brainstem compression (Figure 2).

History of past illness

The patient had no history of systemic diseases.

Physical examination

Neurologic examination revealed a Glasgow Coma Scale score of 8, with no eyes open. The pupils dilated to 4 mm on the right and 2 mm on the left, with left hemiplegia.

Laboratory examinations

On the 7th day after the operation, the patient underwent a blood cell analysis and examinations of blood electrolyte, liver function, and renal function. The test results were all normal, except for a slight decrease in the blood sodium concentration (130.3 mmol/L) and chlorine concentration (94.3 mmol/L). On the 8th day after the operation, examination of blood electrolyte showed sodium concentration (132 mmol/L) and chlorine concentration (95.6 mmol/L).

Imaging examinations

An urgent head CT revealed increasing midline shift, transtentorial herniation, and brainstem compression (Figure 2).

FINAL DIAGNOSIS

The patient was ultimately diagnosed with paradoxical herniation.

TREATMENT

Aggressive medical treatment was immediately initiated to lower intracranial pressure (ICP), including 30° Fowler's position and 20 mg intravenous bolus of furosemide. However, neurological symptoms continued to worsen, and the patient went into lethargy. During the active preparation for the operation, a significant phenomenon was found: Regular brain beats could be seen under his obviously sinking flap. Therefore, we considered that the brain herniation was caused by the brain sag due to low ICP, so we dropped the plan of reoperation. The patient was therefore placed in a supine position and quickly replenished 9% saline intravenously.

OUTCOME AND FOLLOW-UP

A few hours later, with the sinking skin flap relieving, the neurological function of the patient was significantly improved, and consciousness returned. But the left side remained hemiplegia, and the right pupil dilated to 4 mm. In the next day, the head CT showed resolution of transtentorial herniation, a significant decrease in midline shift, and reappearance of basal cisterns (Figure 3A and B). The neurological function of the patient continued to improve, and the patient was successfully changed from a supine position to the Fowler's position within a few days. Two weeks later, he was able to stand and walk with the help of others. The right pupil contracted to 3 mm and the left pupil 2 mm, with a restored light reflection. The head CT shows that hematoma and edema have been absorbed entirely (Figure 3C and D). He was eventually transferred to a rehabilitation facility, waiting for the next skull repair surgery. The 2-mo follow-up revealed a good prognosis with mild hemiplegia on the left side (muscle strength grade 4).

DISCUSSION

Paradoxical herniation, a rare complication of patients who have undergone craniotomy, refers to transtentorial herniation in the context of intracranial hypotension^[3,6]. This life-threatening complication has been reported after various types of CSF depletion and drainage, including CSF leakage, lumbar puncture, lumbar drain, CSF shunt, and ventriculostomy in patients undergoing a decompressive craniectomy^[4,9-11]. The pathological mechanism of this rarely reported complication is unclear and may be related to atmospheric pressure, cerebral gravity, positional changes, and CSF exhaustion^[3,6,7]. Interestingly, there are many reports of sinking skin flap syndromes after large craniectomy, a series of symptoms including motor dysfunction, cognitive dysfunction, headache, mood disorders, and sensory disturbance, most of which can be improved by cranioplasty^[12,13]. The syndromes have a similar pathological mechanism with paradoxical herniation. The difference is that the

symptoms are mild, slow to deteriorate, not life-threatening, and usually occur several months after decompressive craniectomy^[14, 5].

So far, there are only a few cases of paradoxical herniation without CSF drainage. If a patient rebleeding after intracranial hematoma removal, reoperation should be performed to remove the hematoma and cranioplasty; if paradoxical herniation occurs after surgery, the possibility of CSF loss during multiple operations should be considered^[8]. Another report was that paradoxical herniation occurred 1 h after removing acute subdural hematoma. It was suspected that a large amount of CSF was lost during the operation, which was improved by immediate cranioplasty^[6]. Interestingly, a patient with massive cerebral infarction caused by middle cerebral artery embolization developed paradoxical herniation 8 days after decompressive craniectomy, and the neurological function in the upright position continued to deteriorate. These symptoms were resolved by Trendelenburg and hydration quickly, considering the location changes^[7]. Our case is unique in that paradoxical herniation occurred after a rapid intravenous drip of 50 g of 20% mannitol in the Fowler's position, without CSF drainage. Importantly, we found that the skin flap significantly sank after mannitol. For the paradoxical herniation, the patient's life was quickly saved by changing the position and avoiding reoperation in the short term. Interestingly, for increased ICP, several hours after the treatment, including supine position, intravenous rehydration, and stopping mannitol, the depressed flaps gradually protruded with the improvement of the nerve function. It is indicated that the paradoxical herniation is caused by brain sag in this case. Possibly, the dura mater loses the close attachment to the skull, and the brain sags with the effect of its own gravity and the intracranial and extracranial pressure difference. Besides, patients with low flap pressure cannot use large doses of mannitol for a long time, which can lead to life-threatening paradoxical herniation.

Treatment of paradoxical herniation should include immediately placing the patient in the supine or Trendelenburg position, providing intravenous fluids, and discontinuing all medications designed to reduce ICP^[7,8]. In addition, by clamping all

CSF drainage tubes or performing CSF leakage repair to solve the underlying cause of CSF loss, clinical improvement is usually expected within a few hours^[4]. Perhaps emergency cranioplasty is also a treatment option^[6].

CONCLUSION

After large bone craniectomy and decompression, long-term treatment with large amounts of mannitol may cause a rare paradoxical herniation. Paradoxical herniation should be considered if flaps are severely sunken, the brain beating under the skin flap is normal, and head CT excludes secondary intracranial hypertension. Placing the patient in the supine or Trendelenburg position, along with rehydration therapy, may quickly save lives without the need for another operation.

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SIMILARITY INDEX

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