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Recovered bone flap tilting after encephalo-myo-synangiosis: A complication

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ABSTRACT

Objectives: The aim of this study was to analyze the clinical data concerning the loosening and tilting of the recovered bone flap after encephalo-myo-synangiosis (EMS) and explore the potential underlying mechanisms.
Patients and methods: Clinical data of all patients who underwent an EMS procedure from January 1, 2014 to April 30, 2018 at our hospital were collected. The cases with postoperative tilting of the recovered bone flap were identified and the clinical characteristics of the patients involved were analyzed.
Results: There were totally 204 patients who underwent EMS, among whom 12 patients (Male/Female = 6/6) experienced tilting of the recovered bone flap after craniotomy. The average age was 44.3 years. The average height of the tilt was 8.3 ± 3.1 mm. The shortest time for the bone flap to tilt to a height of 5 mm was found to be one day after surgery and the longest was up to 7 months. In cases in which maximum height was observed in a short duration after surgery, the average time for the tilted bone flap to reach the maximum height was 6.1 ± 3.6 days. In addition, a second craniotomy on the opposite side might be an important factor, and three cases were noted with second surgeries (Cases 2, 7, and 9).
Conclusion: Recovered bone flap tilting after EMS should be listed as a complication after roofing of the temporal muscle. This complication may be related to the temporal muscle edema, the increase of intracranial pressure and no bandage compression.

1. Introduction

Encephalo-myo-synangiosis (EMS) is an operation method commonly used to treat Moyamoya disease (MD), also known as abnormal basilar vascular network hyperplasia, which is a chronic occlusive cerebrovascular disease characterized by stenosis or occlusion of distal internal carotid artery and anterior and middle cerebral arteries, and compensatory dilation of perforating arteries at the base of the brain [1]. In MD, the inner layer of the carotid artery proliferates within the arterial lumen. The artery also fills with blood clots, which may cause strokes. In children it tends to cause strokes or seizures. In adults it tends to cause strokes or bleeding.

Since MMD tends to affect only the internal carotid artery and nearby sections of the adjacent anterior and middle cerebral arteries, surgeons can direct other arteries, such as the external carotid artery or the superficial temporal artery to replace its circulation. The arteries are either sewn directly into the brain circulation, or placed on the surface of the brain to reestablish new circulation after a few weeks [2]. Because MD always causes recurrent cerebral ischemia, EMS is thought to be capable of directly supplying blood flow for the ischemic brain tissue through temporal muscle vessels, which could improve the symptoms.During EMS, the temporal muscle was attached to the surface of ischemic brain tissue and then covered with the bone flap [3].

In addition, EMS combined with direct bypass surgery such as superficial temporal artery and middle cerebral arteryanastomosis(STA-MCA) may be more advantageous [4]. In our hospital, we place the temporal muscle on the surface of the brain, and then cover the bone flap. The bone flap would saw a rectangular hole so that the temporal

Abbreviations: MD, Moyamoya disease; EMS, Encephalo-myo-synagiosis; STA-MCA, superficial temporal artery to middle cerebral artery; MCA, middle cerebral artery; CT, computed tomography; DSA, digital subtraction angiography; MRI, magnetic resonance imaging; MRA, magnetic resonance angiography; CTA, computed tomography angiography; MRP, magnetic resonance perfusion; ICA, internal carotid artery; IVH, intraventricular hemorrhage; ACA, anterior cerebral artery; ICP, intracranial pressure

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muscle passes through the skull. If the arteries are suitable for bypass, STA-MCA would be perform meanwhile in most of time.

In recent years, owing to the improvements in fixation materials, loosening of skull bone flaps after craniotomies is currently rarely observed. In particular, there were few reports focusing on the tilting of the recovered bone flap after surgery.However, we observed some cases in recent years where the recovered bone flap loosens and tilts after surgery, especially after EMS.As a result, we have summed up these cases, and in view of this problem, preliminary research and discussion on operation mode, time and possible reasons were made.

2. Ethical review

This observational study was approved by the clinical ethics committee of the Second Affiliated Hospital of Zhejiang University School of Medicine. Informed written consent was obtained from the patients.

3. Patients and methods

We checked the cases of patients who underwent EMS procedures between January 1, 2014 and April 30, 2018, and there were totally 204 patients who underwent EMS, among whom 12 cases with tilting of the recovered bone flap after craniotomy. We realized that this condition has not been well studied before, and therefore, we collected the data of these cases for an analysis and discussion. In this study, all imaging examinations were conducted according to the clinical situation and the images were measured using our hospital's imaging system-Maroland m-viewer and its own measuring tools. In the head computed tomography (CT) image of the patients after the craniotomy, parallel lines were made on the tangent position. The distance between the two parallel lines was defined as the height of the bone flap tilt. We determined a 5-mm height of the bone flap tilt as a positive target (Fig. 1). When patients have a tilted bone flap, the scalp is tight and rough. Sometimes a floating skull can be touched, which always caused a severe tension (Fig. 2a and b). To further study this series of cases, we measured the long diameter and width of the surgical bone window, and recorded the method of fixing the skull and the number of fixed points.

4. Results

In all, 204 cases of EMS surgeries performed at our hospital between January 1, 2014 and April 30, 2018 were included in the study; of these, we found 12 cases showing tilting of the recovered bone flap after roofing of the temporal muscle (Table 1).

Fig. 1. The method of measurement. The parallel lines were made on the tangent position. The distance between the two parallel lines was defined as the height of the bone flap tilt. We determined a 5-mm height of the bone flap tilt as a positive target.

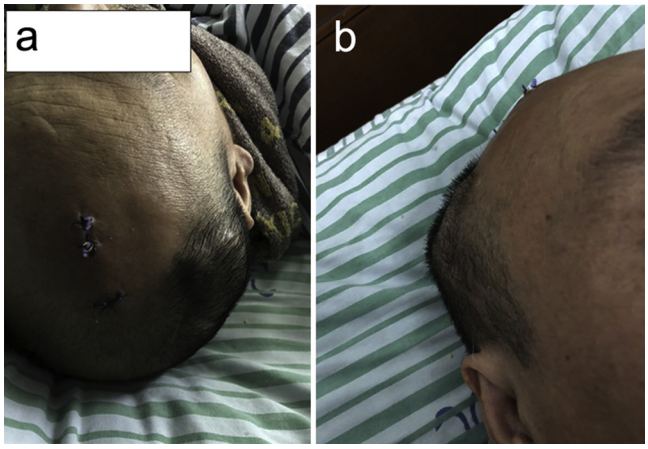

Fig. 2. The scalp of the patient was tight and rough (a); a floating skull can be touched (b).

There are six male patients with an average age of 46.7 years in this series of cases, while six female patients with an average age of 41.8 years; there was no difference between genders. All of the 12 patients did not have osteoporosis. There were 33 patients in the rest of patients in our study having osteoporosis. The minimum height of the tilted bone flap was 5 mm and the maximum was 15 mm. The average height was 8.3 ± 3.1 mm. The shortest duration for the bone flap to uplift to 5 mm was found to be one day after surgery and the longest was up to seven months.

In cases in which maximum height was observed in a short duration after surgery (within one month for Cases 3, 4, 5, 6, 8, 10, 11, and 12), the duration for the tilted bone flap to reach the maximum height was 4, 7, 2, 13, 2, 8, 6, and 7 days after surgery, respectively, with an average of 6.1 ± 3.6 days after surgery. In addition, a second craniotomy on the opposite side might be an important factor, and three cases were noted with second surgeries (Cases 2, 7, and 9). The characteristics of all the cases were summarized in Table 1. The proportion of occurrence of bone flap tilting was found to be 5.88% (12/204). Thus, we believe this should be listed as a complication after roofing of the temporal muscle.

The details of 12 cases are described below:

Case 1. A 46-year-old man who experienced a sudden headache and an intraventricular hemorrhage (IVH) was tested via CT immediately (Fig. 3a). Subsequently, a digital subtraction angiography (DSA) was performed and Moyamoya disease (MD) was diagnosed with a right posterior cerebral artery aneurysm (Fig. 3b–d). Embolization for the aneurysm was performed after 45 days (Fig. 3e–g). After 50 days, we roofed the temporal muscle and superficial temporal artery of the right side to the surface of the brain. We fixed the bone flap using two titanium chains and four titanium screw; however, tilting of the recovered bone flap was observed after three days of the surgery, with a height of 8 mm (Fig. 3h). After 10 months, the patient experienced another IVH (Fig. 3i), and the height of the tilt increased to 15 mm above the bone window (Fig. 3j).

Case 2. A 40-year-old man was diagnosed with MD after an IVH and an aneurysm in the left middle cerebral artery (MCA) (Fig. 4a–c). Half a year later, he presented at our hospital for surgical treatment, and we performed a superficial temporal artery to middle cerebral artery (STA-MCA) bypass and EMS on the right side. No tilting of the bone flap was observed on the second day after surgery (Fig. 4d). Five months later, only a 1-mm rise in the bone flap was observed (Fig. 4e). After a further two months, clipping was performed for an aneurysm, and the bone flap on the right side tilted to a height of 8 mm (Fig. 4f) at two days after this

Table 1
Characteristics of all cases included in the study.

No. of cases	Gender	Age (year)	Bone window (length*width)(cm)	By-pass	No. of Fixed points	Method of fixing	Time to reach uplift 5mm	MH (mm)	Time to reach MH	Other factors
1	M	46	5*5	N	2	titanium chain*	3 days	15	10 months	intraventricular hemorrhage
2	M	40	6*8	Y	3	titanium chain	7 months	8	7 months	opposite side operation
3	M	58	7*8	Y	3	cranial lock*	2 days	11	4 days	
4	F	57	6*9	Y	3	cranial lock	7 days	11	7 days	
5	F	55	6*8	Y	3	cranial lock	2 days	6	2 days	intracerebral hemorrhage
6	F	47	7*9	N	3	titanium chain	3 days	8	13 days	epidural hematoma
7	M	53	6*6	Y	3	titanium chain	75 days	5	75 days	opposite side operation
8	F	30	6*8	Y	3	titanium chain	2 days	11	2 days	
9	F	42	6*7	Y	3	titanium chain	6 months	6	6 months	opposite side operation
10	M	63	5*8	Y	3	titanium chain	8 days	6	8 days	subdural hematoma
11	F	20	6*9	Y	3	titanium chain	6 days	5	6 days	
12	M	50	6*10	Y	3	titanium chain	1 day	7	7 days	

Abbreviations: M, male; F, female; MH, maximun height; N, none; Y, yes.
*titanium chainthis means use two titanium nails to fix a titanium chain with two holes, in which one head of the titanium chain fix on the bone flap and another head fix on the skull around the bone window.
*cranial lock (CranioFix® absorbable in trade name) is a kind of absorbable cranial fixation system made by B.Braum (https//www.bbraun.com/en/products/b0/craniofix-absorbable.html), in which two pieces of polymer material were fixed with silk thread on both side of the bone flap.

surgery.

Case 3. A 58-year-old man had repeated dizziness and headache for one year. DSA conducted at a local hospital confirmed MD. The left internal carotid artery (ICA) was occluded. Subsequently, left-side STA-MCA bypass and EMS were performed at our hospital. CT conducted on the second day after surgery suggested that the bone flap was tilted by about 10 mm (Fig. 5a and 5b). CT scans on the fourth day after surgery showed a tilt height of about 11 mm (Fig. 5c).

Case 4. A 57-year-old woman presented with numbness in her left hand for one year, aggravated with memory loss. A head magnetic resonance imaging (MRI) and CT showed multiple cerebral infarctions (Fig. 6a and b). DSA suggested MD (Fig. 6c and 6d) and the patient underwent a right-side STA-MCA bypass and EMS. No tilting of the bone flap was observed on the second day after surgery (Fig. 6e), but an 11-mm tilt was observed (Fig. 6f and 6 g) on the seventh day after surgery, which was still evident at two weeks (Fig. 6h).

Case 5. A 55-year-old woman had a sudden fall and a hemorrhage was diagnosed in the left ventricular antrum (Fig. 7a). DSA (Fig. 7b–d) was performed and was suggestive of MD. Further, occlusion of the left ICA and posterior circulation vascular compensation were observed. After four months, a left-side STA-MCA bypass and EMS were performed, and CT conducted two days after surgery showed bone flap tilting of about 6 mm (Fig. 7e and f) and minor intracerebral hemorrhage near the operation area (Fig. 7g-h).

Case 6. A 47-year-old woman experienced sudden syncope. She was diagnosed with MD, an anterior choroidal artery aneurysm, and occlusion of the right MCA. After 12 days, right-side STA-MCA bypass, EMS, and right anterior choroidal artery aneurysm clipping were performed at our hospital. DSA after surgery showed an open bridge vessel and left anterior cerebral artery (ACA) and MCA stenosis (Supplemental Fig. 1a–c). Subsequently, eight months after surgery, CT showed no shift in the bone flap (Supplemental Fig. 1d), and roofing of temporal muscle on the left side was then performed. Bone flaps on both sides were in place immediately after surgery (Supplemental Fig. 1e), and after 3 days, CT showed that the bone flap on the left side was tilted by about 5 mm (Supplemental Fig. 1f). Eight days after surgery, an epidural hematoma was found on the distant left side of the

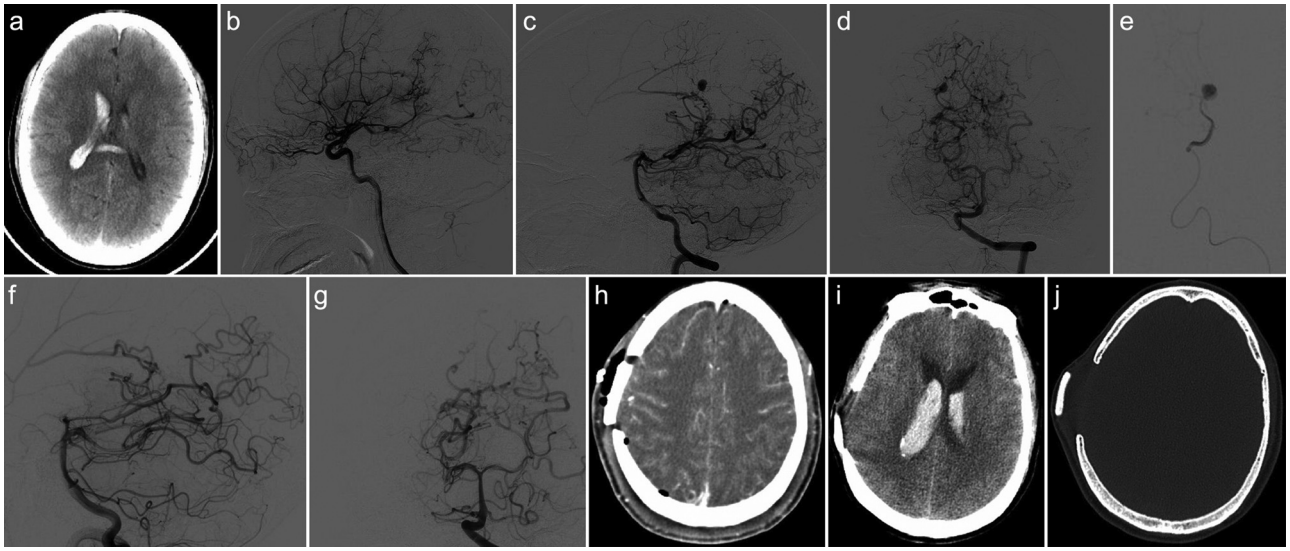

Fig. 3. The images of Case 1: The CT scan before operation showed the IVH (a); DSA showed MD with a right posterior cerebral artery aneurysm (b–d); embolization for the aneurysm (e–g); tilting of recovered bone flap three days after surgery with a height of 8 mm (h); recurrent IVH after 10 months (i); the bone flap height increased to 15 mm above the bone window (*j*).

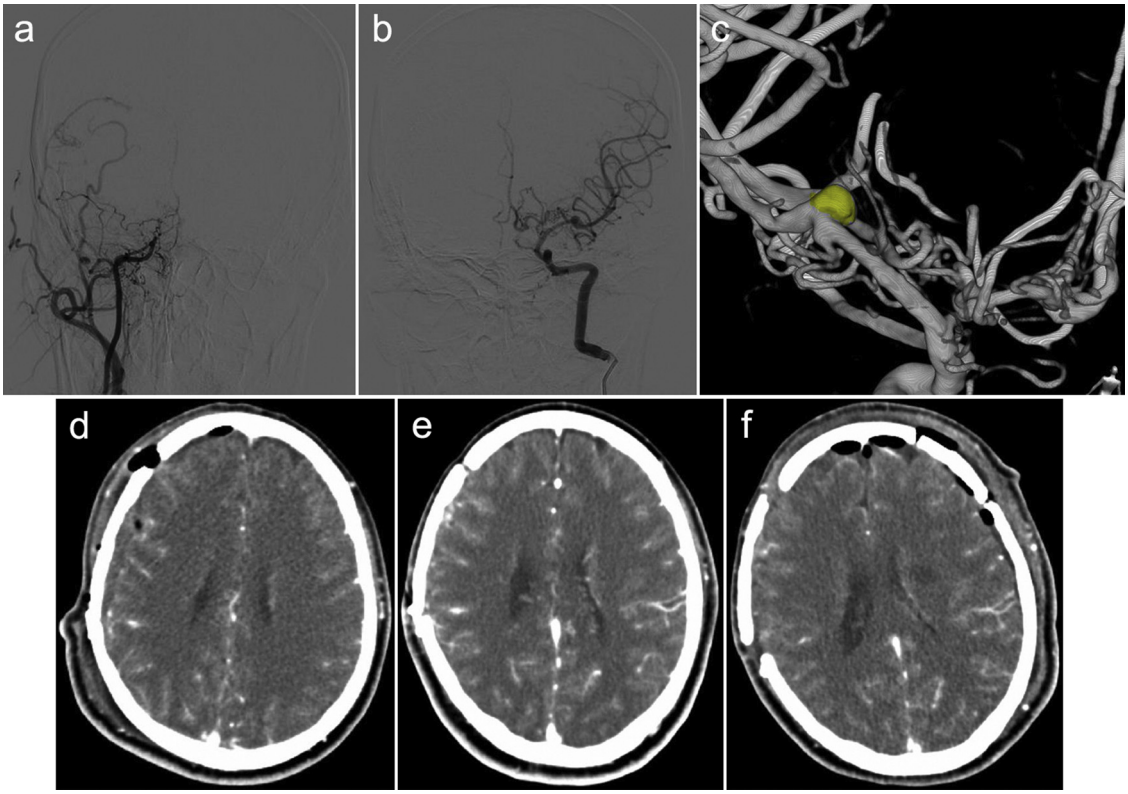

Fig. 4. The images of Case 2: DSA showed MD and an aneurysm in left MCA (a–c); the bone flap was not tilted at the second day after surgery (d); the bone flap tilted with a height of 1 mm after five months (e); the bone flap on the right side tilted with a height of 8 mm two days after clipping of the aneurysm (f).

left surgery site (Supplemental Fig. 1g). CT was performed 13 days after the operation, and it confirmed that the bone flap tilt had increased to 8 mm (Supplemental Fig. 1h and i). The epidural hematoma was gradually resorbed 28 days after the operation; the tilting of the bone flap was reduced to about 5 mm (Supplemental Fig. 1j and k).

Case 7. A 53-year-old man felt dizzy for five months. Magnetic resonance angiography (MRA) revealed MCA occlusion (Supplemental Fig. 2a). At our hospital, a right-side STA-MCA bypass and EMS were performed. CT showed slight tilting of the bone flap (about 2 mm) on the third day after surgery (Supplemental Fig. 2b). On the eighth day after surgery, the bone flap tilt was about 5 mm (Supplemental Fig. 2c). The bone flap tilt decreased to about 1 mm 70 days after the surgery (Supplemental Fig. 2d), and this was followed by a EMS surgery on the left side. Two days after this surgery, CT revealed that bone flaps on both sides were tilted and the tilt on the right side returned to 5 mm (Supplemental Fig. 2e).

Case 8. A 30-year-old woman with sudden onset of a headache presented at our hospital. CT showed right thalamic hemorrhage breaking into the ventricle (Supplemental Fig. 3a); hematoma was absorbed after conservative treatment (Supplemental Fig. 3b), and DSA examination confirmed MD (Supplemental Fig. 3c and 3d). Six months later, right-side STA-MCA bypass and EMS were performed, and two days after this procedure, tilting of the bone flap by about 11 mm was noted.

Case 9. A 42-year-old female suffered the right-side limb weakness for half a month. DSA indicated MD; ICA, ACA, and MCA occlusion was reported (Supplemental Fig. 4a and b). A left-side STA-MCA bypass and EMS were performed. CT scan showed bone flap tilting by approximately 3 mm (Supplemental Fig. 4c) 5 days after the surgery. Half a year later, STA-MCA bypass and EMS of the right side were performed, and one day later, CT showed left bone flap tilting of 6 mm (Supplemental Fig. 4d).

Case 10. A 63-year-old man, experienced left limb weakness for half a year. MRA prompted right ICA stenosis (Supplemental Fig. 5a). He was

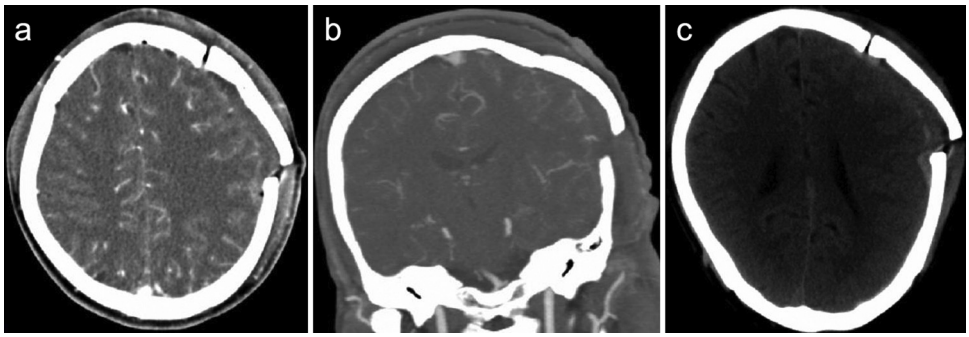

Fig. 5. The images of Case 3: The CT scan showed that the bone flap was tilted with a height of 10 mm two days after surgery (a and b); the bone flap was tilted by about 11 mm four days after surgery (c).

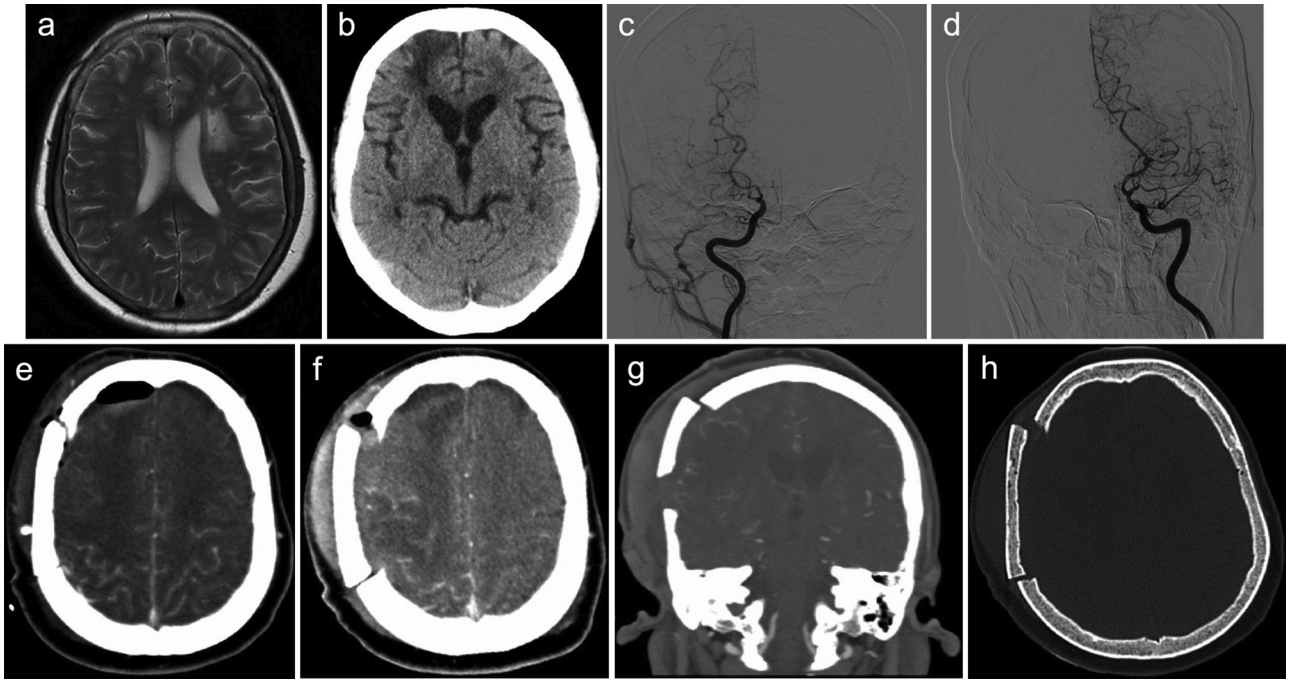

Fig. 6. The images of Case 4: The MRI showed multiple cerebral infarctions (a and b); the DSA showed MD (c and d); the CT scan showed that the bone flap was not tilted on the second day after surgery (e); the bone flap was tilted by about 11 mm on the seventh day after surgery (f and g) and the tilt was still evident after two weeks (Fig h).

treated with drugs but decreased muscle strength in left limbs was noted. Later, computed tomography angiography (CTA) showed right ICA occlusion (Supplemental Fig. 5b), and magnetic resonance perfusion (MRP) suggested right hemisphere hypoperfusion (Supplemental Fig. 5c). STA-MCA bypass and EMS of the right side were performed. Two days postoperatively we observed tilting of the bone flap by about 4 mm with subdural hematoma (Supplemental Fig. 5d), and CT on the eighth day showed a bone flap tilt of up to 6 mm (Supplemental Fig. 5e). After two years and eight months, CT showed that the bone flap was no longer tilted and the subdural hematoma was significantly reduced (Supplemental Fig. 5f).

Case 11. A 20-year-old female showed repeated numbness of right arm for a year. DSA indicated MD, and left ICA, ACA, and MCA stenosis was observed (Supplemental Fig. 6a). Accordingly, left-side STA-MCA

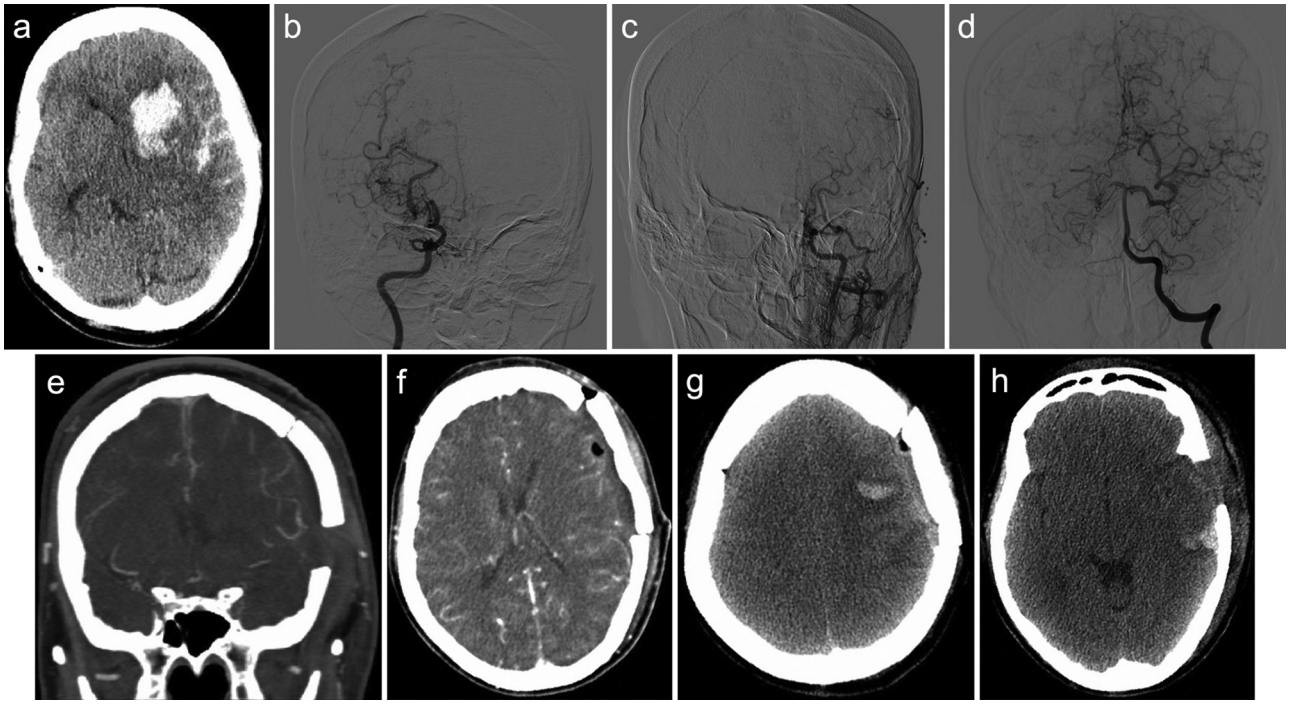

Fig. 7. The images of Case 5: The CT scan showed hemorrhage in the left ventricle antrum (a); DSA showed MD, left ICA occlusion and posterior circulation vascular compensation (b–d); the CT scan showed that the bone flap tilted by about 6 mm two days after surgery (e and f); minor intracerebral hemorrhage near the operation area two days after surgery (g–h).

bypass and EMS were performed. On the second day after surgery, the bone flap tilted by 3 mm (Supplemental Fig. 6b). On the sixth day, CT revealed bone flap tilting of up to 5 mm (Supplemental Fig. 6c).

Case 12. A 50-year-old male experienced right limb weakness and slurred speech for two days. MRI prompted multiple basal ganglia infarctions (Supplemental Fig. 7a), and MRA suggested left ICA and MCA stenosis (Supplemental Fig. 7b). Three weeks later, left STA-MCA bypass and EMS were performed. On the second day after surgery, the bone flap was tilted by about 4 mm (Supplemental Fig. 7c), and increased to 7 mm (Supplemental Fig. 7d) on the seventh day after surgery. The tilting of the bone flap was back to 3 mm (Supplemental Fig. 7e) at two months after surgery, but lifted to 7 mm (Supplemental Fig. 7f) at eight months after surgery.

5. Discussion

We summarized the possible causes of this situation as follows. First, the temporal muscle is placed under the bone flap. Placement of the temporal muscle produces a space-occupying effect. Because of a different thickness of the temporal muscle and its rich blood supply, there will be significant edema in the short term after surgery; strong muscle contractions provide a persistent and non-homogenous force on the bone flap. This may be the main reason for this situation. In cases in which bone flap tilting takes place at an early postoperative stage, the time to reach the maximum height of tilting is 6.1 ± 3.6 days after surgery, which is also the stage when temporal muscle swelling rather serious. Second, postoperative intracranial pressure is usually high. In order to ensure the cerebral perfusion of these patients, postoperative mannitol was not commonly used. However, postoperative brain and muscle edema lead to increased intracranial pressure. Thus, increased pressure may lead to tilting of the bone flaps. Third, incisions are not routinely pressure bandaged. The patients with MD or secondary chronic intracerebral vascular occlusion in this study always underwent STA-MCA bypass (in 10 of the 12 cases, STA-MCA bypass and EMS were simultaneously performed). After the operation, to ensure sufficient blood supply to the bypass vessels, the incision is generally not subjected to conventional elastic bandage compression, making the strength of the bone flap fixation relatively weak, and this makes it likely to move outward because of the increase in intracranial pressure. Fourth, some complications after surgery, a second surgery, or other factors, such as intracranial hemorrhage, edema, and infarction, may cause intracranial pressure fluctuations and immediately increase the pressure, leading to tilting. In our study, 7 of the 12 cases (1, 2, 5, 6, 7, 9, and 10) meet one of the above situations, and at least 6 of them (1, 2, 6, 7, 9, and 10) show a clear relationship of the abovementioned situations with the bone flap tilting. However, we did not observe cases of bone flap tilting in cases of postoperative intracranial hemorrhage or contralateral reoperation that we summarized in other cases of craniotomy.

Some doctors suggested osteoporosis as a possible reason, but the average age of those patients was 46.7 years for men and 41.8 years for women. In addition, none of the 12 patients was diagnosed with osteoporosis. On the contrary, the others out of the 12 patients in our study who had osteoporosis did not suffer from bone flap tilting after surgery. We also proposed that the tilting may have been resulted because of unsecure fixation of the bone flaps. However, this does not seem to be the case as even if we consider this to be a manual error during surgery, all surgeries were performed by the same surgeon and no screws were left loose. Further, there no loosening of other skull fixations was observed. Moreover, similar cases of bone flap tilting did not occur in other types of surgeries performed during the same period.

We also recognized that this tilting of the bone flaps may be a compensatory measure and may be helpful in preventing cerebral compression. Stoner et al. [5] analyzed the volume and height of cerebral herniation in the skull bone window after decompressive craniotomy in a group of patients with ischemic stroke, and proposed that after the craniotomy, the bone flap should be repositioned by 5 mm to prevent bone flap decompression surgery. Some studies on hinged bone flaps and floating bone flaps also support the use of such techniques during brain injury, hemorrhage, cerebral infarction, and narrow cranial disease [6–9]. Considering that in four cases in our group, IVH, epidural hematoma, subdural hematoma, and intracerebral hematoma occurred after surgery and in another 3 cases, operations were performed on the contralateral side subsequently, the loosening and tilting of the bone flap may be precisely because of an increase of intracranial pressure in a short time after surgery, and it is precisely a protective mechanism that reduces compression and damage to the brain tissue.

There were no reports concerning the solution after skull flap tilting. Early correction or delayed deformity might be helpful. Increase of the number of titanium chains might make the skull more stable and reduce the incidence of skull flap tilting. In fact, we recently tried increasing the fixation point to improve the effectiveness of bone flap fixation, we will continue to pay attention to the follow-up of this part of patients. There are also some helpful suggestions worthy of absorbing and trying: In cases where temporal muscle is thick, temporal muscle should be made relatively thin and bone flap should be drilled in order not to press the brain. If the intracranial pressure is too high after EMS, it is necessary to perform craniotomy, but it is necessary to pay attention to the protection of the bridge blood vessels.

6. Limitations

As this study included few numbers of cases and was a single-center study with a single medical team, more data are needed to assess whether this complication requires positive correction or current surgical skills for EMS need to be properly improved. This series of cases presents a complication that is rare and needs attention and further research.

Mainly due to economic reasons and lack of cranial fixation system could be chosen, as meanwhile, didn't consider of such complications, we have almost all adopted two or three-point fixation (three titanium chains + six titanium nails or three pieces of "cranial lock"). Titanium chains and "cranial lock" are soft, their force area are small and the strength is insufficient, at the same time, three fixation points are sometimes not enough. Some experts have suggested that the occurrence of such bone flaps is mainly due to the fact that the fixation method is not stable enough, titanium plate fixation can effectively prevent this complication from happening.But there is a fact that the price of a titanium plate is much higher than titanium chains, and fixing the titanium plate will require more titanium nails. We will select suitable cases to try the fixation with titanium plate in future and follow-up. Despite the many limitations and shortcomings, we believe that this is a rarely reported complication of a series of cases with similar characteristics that are worth sharing and further discussion.

7. Conclusion

Recovered bone flap tilting after EMS should be listed as a complication after roofing of the temporal muscle. This complication may be related to the temporal muscle edema, the increase of intracranial pressure and no bandage compression.

Conflict of interests

The authors have no conflict of interests to declare.

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None.

Acknowledgments

None.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.clineuro.2019.01.009.

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