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A Case of Cavernous Malformation of the Midbrain Removed via an Interhemispheric Transcallosal Subchoroidal Approach

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Abstract

Cavernous malformations of the midbrain have a higher rate of hemorrhage and a poorer prognosis than vascular malformations of other brain areas. Surgical resection of these lesions is often necessary to avoid neurological deficits in affected patients. Herein, the literature surrounding cavernous malformations was examined, and the case of a 48-year-old man with left hemiparesis and diplopia caused by incomplete right oculomotor nerve palsy, who was diagnosed with a hemorrhage from a midbrain cavernous malformation, was discussed. The lesion expanded gradually on magnetic resonance imaging and was symptomatic; radical removal of the lesion before the onset of irreversible symptoms due to recurring bleeding was therefore considered to be beneficial for the patient. Surgical removal of the entire cavernous malformations of the midbrain was performed using an interhemispheric transcallosal subchoroidal approach, with excellent postoperative results and complete recovery from the oculomotor nerve palsy and left hemiparesis. This case shows that this approach is the most appropriate for surgical resections of lesions in the upper midbrain.

Keywords: cavernous malformation, midbrain, intracranial hemorrhage, oculomotor nerve palsy, interhemispheric transcallosal subchoroidal approach

Introduction

Cavernous malformations of the brainstem (CMBS) have a higher hemorrhage rate and poorer prognosis compared to cavernous malformations of other brain regions.[1,2] In symptomatic CMBS as well as cases with increasing lesion size, treatment via resection and gamma knife radiosurgery (GKRS) is considered.[3] When considering direct surgery for CMBS, several important factors must be considered in an attempt to avoid neurological deficits as a consequence of CMBS's trajectory. First, the lesion must be approached from the safe entry zone, and, second, the lesion must be approached from the region where it is most exposed to the surface of the brainstem. This is termed the two-point method.[4] Herein, a case of symptomatic CMBS that was removed via an interhemispheric transcallosal subchoroidal approach based on the two-point method is reported, and an excellent postoperative course, accompanied by a review of the associated literature, is shown.

Case Report

History of present illness

A 48-year-old man suffering from diplopia as a result of incomplete right oculomotor nerve palsy was diagnosed with a hemorrhage of a midbrain cavernous malformation (CM). The patient was subsequently referred to the department of the current study for further investigation and treatment.

Preoperative neurological findings

The patient presented with clear consciousness, right oculomotor nerve palsy, and mild insufficiency of the left hemiparesis. Due to oculomotor nerve palsy, the patient presented with slight abduction of the left eye, as well as impaired adduction, supination, and inversion disturbances

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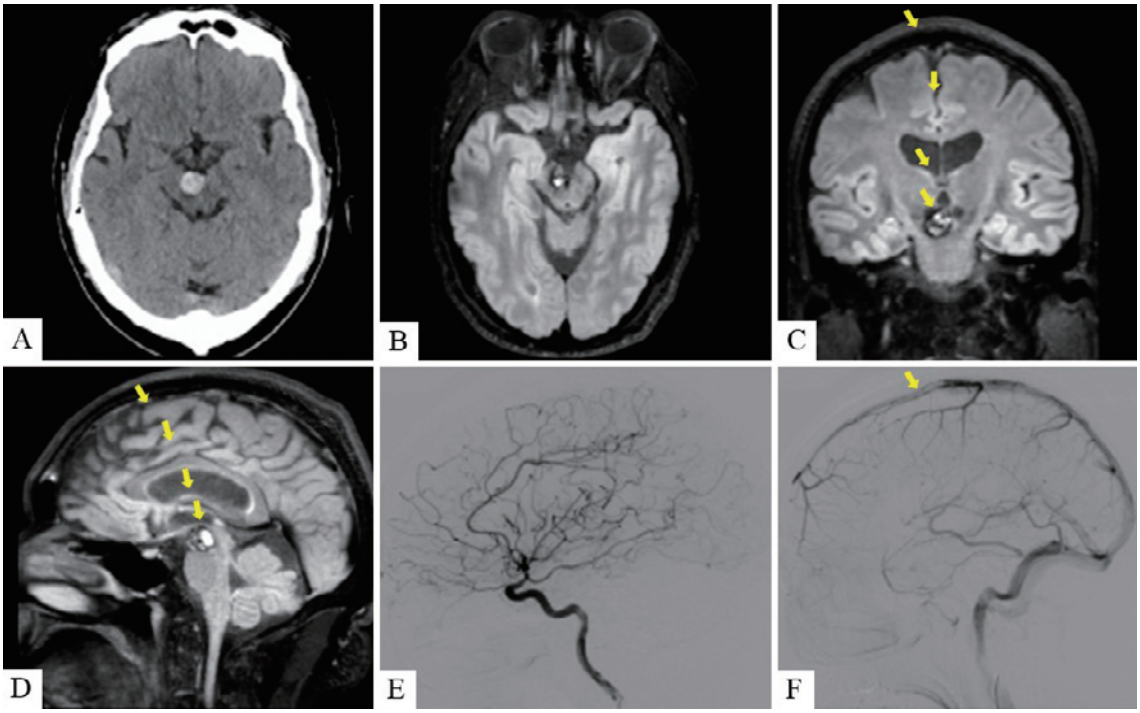

Fig. 1 Preoperative images. A Computed tomography scan of the high-density area slightly right and lateral to the midline of the midbrain. B FLAIR axial image showing the cavernoma on the right side of the midbrain. C and D The *yellow arrows* indicate the approach line for the midbrain cavernoma resection. E and F Cerebral angiography showing no abnormal vascularity and suitable bridging veins for the right interhemispheric approach. The *yellow arrows* indicate the approach line.

of the right eye, mild ptosis, and a sluggish indirect counter reflex.

Preoperative neuroimaging

Computed tomography (CT) scans revealed a high-density area slightly lateral to the midline of the midbrain on the right side (Fig. 1A). Magnetic resonance imaging (MRI) revealed a mass lesion with a hemosiderin rim in the same region (Fig. 1B-D). Cerebral angiography revealed no vascular malformations, including venous malformations and bridging veins suitable for the interhemispheric approach (Fig. 1E, F).

Definition of approach

The CMBS was expanding gradually on MRI and was symptomatic; radical removal of the lesion was therefore considered to be beneficial for the patient before the onset of irreversible symptoms caused by recurring bleeding from CM. The CMBS was located 2 mm to the right of the midline of the midbrain and was in contact with the floor of the third ventricle. The right interhemispheric transcallosal subchoroidal approach was therefore considered to be the most appropriate course of action due to its minimally invasive nature.

Operative findings

The patient was placed in the supine position under general anesthesia and monitored using motor-evoked potential (MEP) and somatosensory-evoked potential (SEP). After determining the optimal approach route using the navigation system, the interhemispheric fissure was divided to expose the corpus callosum between the bilateral pericallosal arteries. A 10-mm corpus callosum incision was made to enter the right ventricle (Fig. 2A). The Monroe foramen, anterior transparent septal vein, choroid plexus, and thalamostriate vein were identified in the lateral ventricle (Fig. 2B). The third ventricle below the choroidal fissure was then entered to expose the floor of the third ventricle (Fig. 2C, D). The CM was not immediately visible on the floor of the third ventricle and no color change was observed. A vertical incision was made 2 mm to the right of the midline, based on preoperative MRI and anatomy of the third ventricle floor, and confirming the hematoma location using the navigation system. Following this, an old fluid hematoma leaked, which was aspirated and decompressed (Fig. 2E, F). The main body of the CM was identified and fully resected without removing the hemosiderin rim (Fig. 2G, H). Pathological findings were consistent with a diagnosis of CMBS. During surgery, MEP and SEP were stable.

Postoperative course

The right oculomotor nerve palsy and left hemiparesis did not worsen; however, the patient did present with cog-

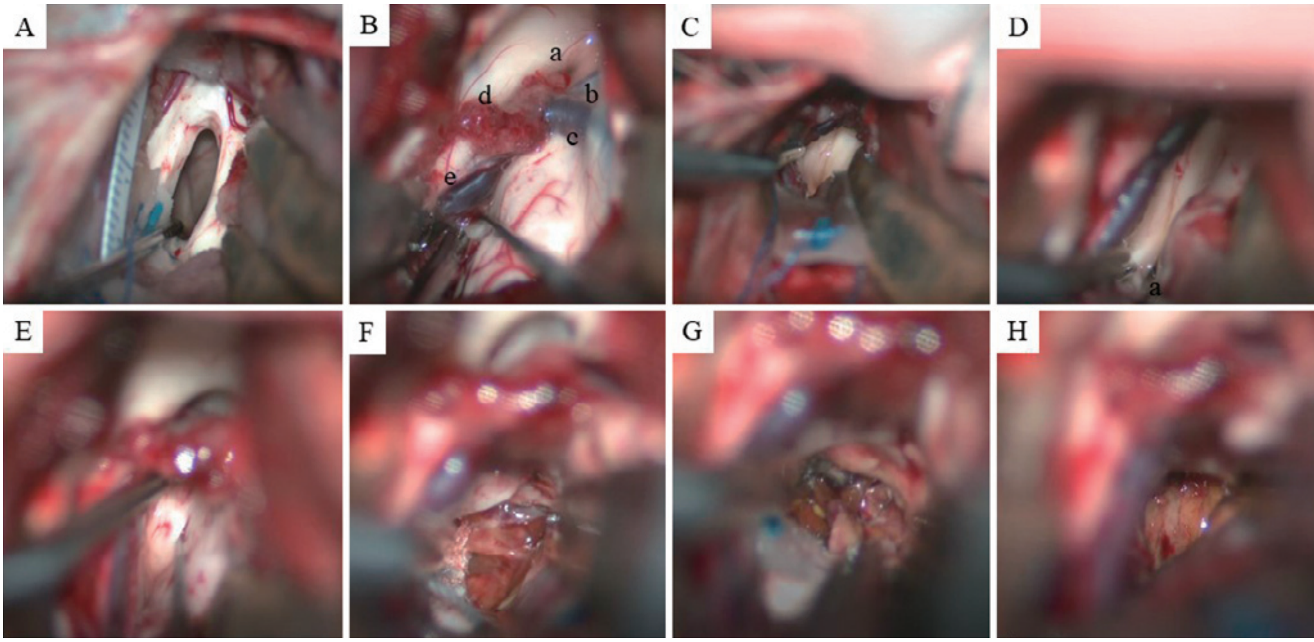

Fig. 2 Intraoperative findings. A The interhemispheric fissure was divided to expose the corpus callosum between the bilateral pericallosal arteries, and a 10-mm incision in the corpus collosum was made to enter the right ventricle. B The view in the right lateral ventricle where *a* Monroe foramen, *b* anterior transparent septal vein, *c* thalamostriate vein, *d* choroid plexus, and *e* internal cerebral vein. C and D The floor of the third ventricle observed from the subchoroidal approach. *a* Aqueduct. E The vertical incision 2 mm to the right of the midline. F An old fluid hematoma that leaked and was aspirated and decompressed. G and H The main body of CM was identified and grossly removed completely without removing the hemosiderin rim.

nitive dysfunction such as short-term memory disturbance, executive dysfunction, and attention dysfunction. The preoperative Mini-Mental State Examination (MMSE) was a perfect score of 30, but it decreased to 21 after the operation. These symptoms improved gradually 4 weeks postoperatively. Postoperative CT and MRI showed that the CM was fully resected. However, MRI, diffusion-weighted image (DWI) and fluid-attenuated inversion recovery (FLAIR) showed high intensity in the corpus callosum and right anterior thalamus (Fig. 3). No cerebral infarction was present. One month postoperatively, the lesions in the corpus callosum and right thalamus disappeared and were no longer visible on an MRI with the patient's symptoms improving (Fig. 4). The patient's cognitive function was fully restored (MMSE score was the perfect score of 30) and oculomotor nerve palsy had recovered 3 months postoperatively.

Discussion

Indication of surgery for symptomatic midbrain cavernous malformations

The rate of hemorrhage and rebleeding in CMBS is higher than that of CMs in other parts of the brain.[1,2] The annual hemorrhage rate and permanent neurological morbidity are notably high in CMs of the brain stem, thalamus, and basal ganglia. The reduction of hemorrhage risk after stereotactic radiosurgery for CMs, especially when located within the parenchyma of the brain stem or diencephalon, is discussed in this literature review. In many reports, the protective role of propranolol administration in preventing hemorrhages of CMs or shrinking of CMs is discussed. The predictors of persistent oculomotor palsy are the time interval between symptom onset and surgery as well as the patient's age. Patients >40 years old are at a greater risk of persistent oculomotor palsy, and early surgery is therefore recommended in patients with oculomotor disturbances.[5] It takes time for medical treatment and GKRS to be effective. Surgical excision is the most common intervention for CMBS, but GKRS is increasingly used. Excision is often the treatment of choice if the lesion is in direct contact with the surface of the brainstem or is considerably large. The gamma knife is often selected if the lesion is located deep within the brainstem or is small.[6] Conversely, a deep intrinsic midbrain CMs location is not necessarily associated with an unfavorable clinical outcome after microsurgical lesionectomy.[7] In this case, the size of the lesion increased on imaging due to repeated hemorrhaging, which resulted in oculomotor nerve palsy and paralysis. Therefore, direct microsurgical treatment was performed.

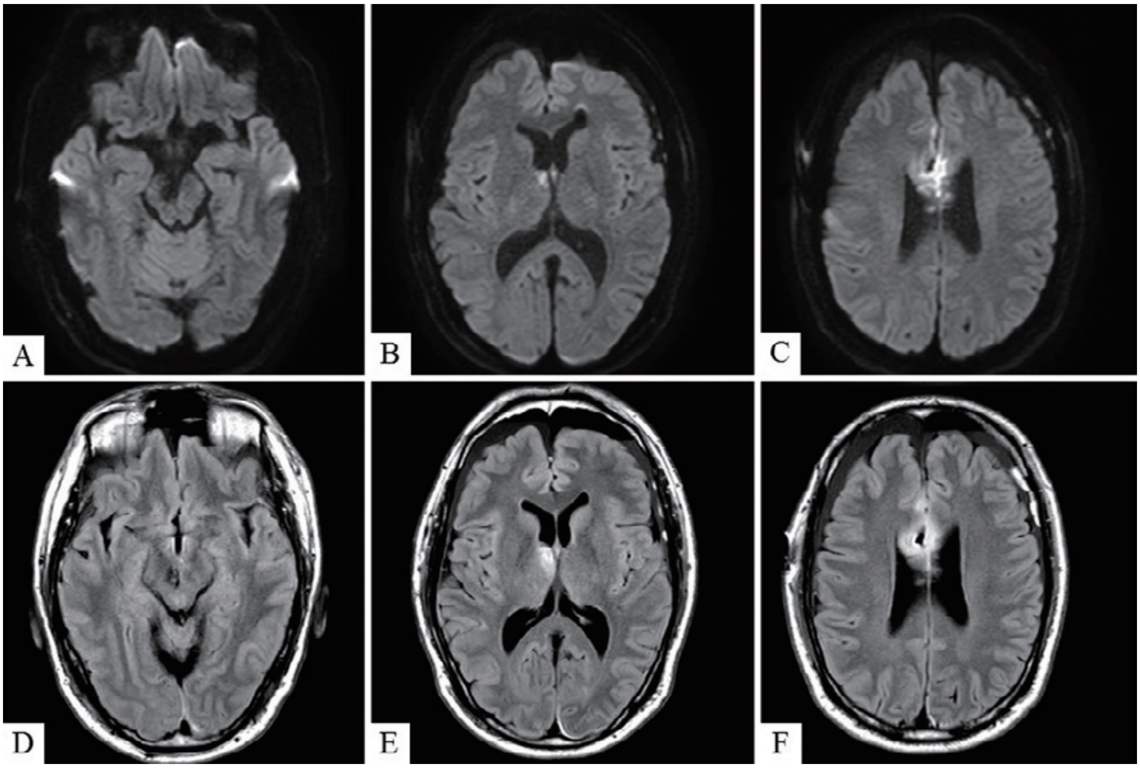

Fig. 3 Postoperative images. A–C Postoperative DW MRI showing a high-intensity area around the corpus callosum and right anterior thalamus. D–F Postoperative MRI FLAIR showing the same findings as DWI.

Choice of approach for midbrain cavernous malformations

The most common strategy for CMBS removal is to approach the lesion from the safe entry zone or the area closest to the surface of the brainstem, termed the two-point method.[4] The safe entry zones for the midbrain were described as the anterior mesencephalic zone, lateral mesencephalic sulcus, and intercollicular region based on anatomical structure. Cavalcanti et al. described an orbitozygomatic or pterional, subtemporal, and median supracerebellar infratentorial approaches to expose the anterior, lateral, and posterior surfaces of the midbrain, respectively.[8] In this case, the CM was located interpeduncular fossa, was exophytic and disrupted the surface of the midbrain surface. Consequently, an ipsilateral or contralateral interpeduncular fossa approach may have been suitable for this case.[9] However, this may have required a combined skull-based approach with a zygomatic arch resection.[10,11] In addition, this approach may be technically challenging due to a narrow surgical field and required the manipulation of important neurovascular structures such as the basilar artery, posterior cerebral artery, superior cerebellar artery, and oculomotor nerve.

An interhemispheric transcallosal transchoroidal or subchoroidal approach was ultimately decided because of the location of the CMBS of the patient in this study. Limited reports were noted on the use of the transcallosal approach for SMBS removal.[12,13] This approach has two major advantages: first, craniotomy is simple and quick, and the midline approach avoids damage to the brain surface.[12,14] Second, the final surgical view is good, and the entire floor of the third ventricle can be observed, even if the lesion extends into the thalamus.[12,14] Conversely, two major points were observed in this approach: first, disconnection syndrome may occur with callosotomy, and callosotomy should be performed at ≤2 cm to prevent this disorder.[15] Second, intraoperative manipulation is deeper than other approaches.[16] Three routes of entry were observed from the lateral ventricles into the third ventricle: the transforaminal, the transchoroidal, and the subchoroidal approaches.[17] The transforaminal approach is associated with a risk of fornix or internal cerebral vein injury and is difficult to use except in cases with a cave of the septum pellucidum. The transchoroidal and the subchoroidal approach are both approaches via the choroidal fissure. The latter has the disadvantage of the risk of thalamic and thalamostriate vein injury compared with the former; however, the risk of fornix and internal cerebral vein injury is lower.[16,18] In addition, when the lesion is localized lateral to the floor of the third ventricle, the transchoroidal approach may be useful as a surgical view for an ipsilateral lesion. However, the lesion was localized slightly lateral to the midline in the current case. Thus, both transchoroidal and subchoroidal approaches were considered useful.[15]

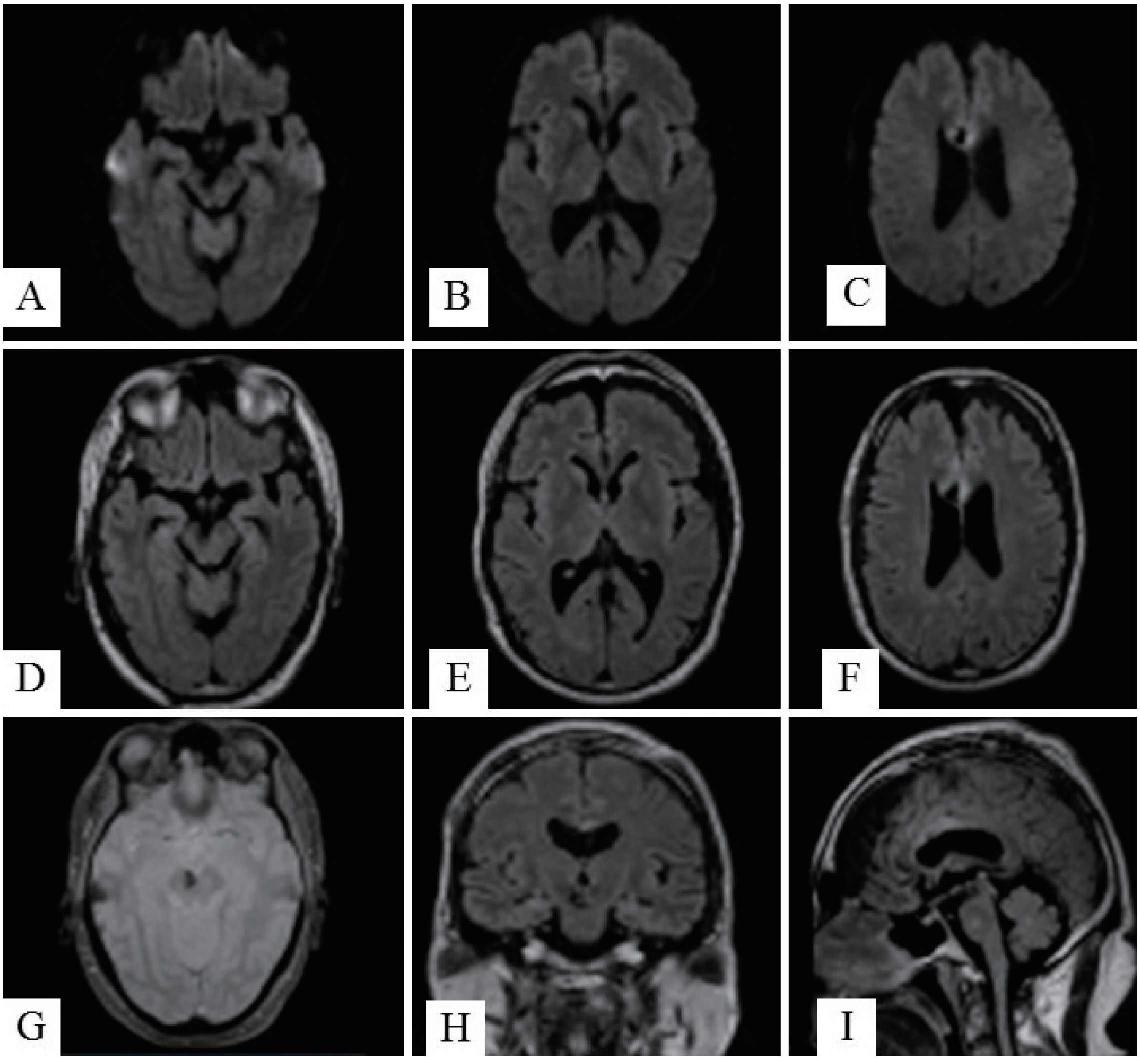

Fig. 4 Magnetic resonance image 1 month after surgery. A-F DWI and FLAIR showing that the hyperintense lesions in the thalamus and corpus callosum disappeared 1 month postoperatively. G-I T2* and FLAIR showing that the cavernoma was completely removed.

Moreover, the fornix becomes thicker posterior to the choroidal fissure, limiting the length that can be opened with a transchoroidal approach. For the above reasons, in the current case, a subchoroidal approach was used to enter the third ventricle through the lateral ventricle to avoid damage to the fornix and internal cerebral vein because the lesion existed in the posterior two-thirds of the third ventricle.[17)]

In the current case, postoperative cognitive dysfunction was observed temporarily 3 weeks after surgery. MRI showed a high-intensity area in the corpus callosum and the right anterior thalamus on DWI and FLAIR, suggesting the influence of compression of the thalamus with a brain spatula, 1 week postoperatively. Even with the transforaminal approach, permanent or transient memory impairment can occur as a complication. The mechanism of this impairment is thought to involve damage to the fornix.[15,19)] In this case, no cerebral fornix damage was observed, as the high-intensity area on the right anterior thalamus disappeared, and the patient's cognitive dysfunction improved. This initial high-intensity area on the thalamus was due to the gentle compression of the medial thalamus to access and remove the CMBS.

The transcallosal subchoroidal approach may be suitable for upper midbrain CMs under the posterior two-thirds of the floor of the third ventricle with careful callosotomy and thalamus compression.

Conclusion

This study reports a case in which a symptomatic CMBS with a tendency to increase over time was removed via the interhemispheric transcallosal subchoroidal approach. Although the surgical field was deep, this approach was useful for the resection of cavernous malformations localized in the upper midbrain. The techniques discussed in this paper, as well as the detailed case report, may give clinicians a better understanding of CMBS and provide an avenue for them to develop the most effective approach for the removal of this pathology should they encounter it themselves.

Ethical Considerations

The authors have no relevant financial or nonfinancial interests to disclose.

This case report has been approved by the Tokyo Women's Medical University Ethics Committee.

The authors obtained the informed consent of this patient.

Conflicts of Interest Disclosure

The authors have no conflicts of interest directly relevant to the content of this article.

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