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Case Report

Sylvian fissure lipoma associated with fusiform aneurysm in the middle cerebral artery trifurcation: A case report and literature review

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ABSTRACT

Background: The intracranial lipomas are rare congenital malformations accounting for approximately 0.1–1.3% of all intracranial tumors, of which Sylvian fissure lipomas account for <5%. These lesions are frequently associated with dysgenesis of neuronal brain tissues and vascular malformations and in the majority are asymptomatic. Intracranial lipomas on magnetic resonance imaging (MRI) may mimic late subacute hemorrhage due to similar radiological features. Due to the tight adhesion of the lipoma to the surrounding nerve structures and vessels, complete removal is difficult and does not guarantee the disappearance of symptoms.

Case Description: We present the case of a 42-year-old woman with chronic headaches and short-term memory impairment who was admitted to the emergency room after an out-of-hospital brain MRI with suspected ruptured right middle cerebral artery (MCA) aneurysm and late subacute intracranial hemorrhage. In the hospital, after clinical evaluation, emergency computed tomography (CT) angiography was performed, which revealed an unruptured fusiform aneurysm located in the right MCA trifurcation surrounded by an extremely hypodense lesion corresponding to fat in the right Sylvian fissure. No features of intracranial hemorrhage were present. The diagnosis of intracranial lipoma was finally confirmed after the MRI of the brain with a fat suppression sequence. Surgical treatment was not attempted, and the patient was treated conservatively with a satisfactory general outcome.

Conclusion: A Sylvian fissure lipoma may be associated with a fusiform aneurysm in the MCA trifurcation. By modifying the standard MRI protocol and performing a CT scan, an intracranial lipoma can be detected and a late subacute intracranial hemorrhage can be excluded.

Keywords: Aneurysm, Intracranial lipoma, Malformation, Middle cerebral artery, Sylvian fissure

INTRODUCTION

Intracranial lipomas are considered sporadic congenital malformations originating from abnormal fatty transformation and maldifferentiation of meninx primitiva at embryonic level.[15,25] The incidence is low and accounts for 0.1–1.3% of all intracranial tumors.[21,22] They are widely distributed in the intracranial spaces with the highest predominance to the midline, situated especially at pericallosal cistern and with exceedingly low prevalence to Sylvian fissure.[6,11,17,25]

These lesions are usually discovered accidentally during diagnostic imaging and unless symptomatic most intracranial lipomas are of doubtful clinical significance.[8,12,23] About half of the cases are associated with specific cerebral conditions, particularly neural structures dysgenesis, obstructive hydrocephalus, and vascular malformations being most relevant.[11,25-27]

In specific MR sequences, lipomas due to its electromagnetic characteristics may mimic extracellular methemoglobin present in late subacute hemorrhage, contributing to challenging diagnostic issues, in particular when intracranial aneurysm is present.[19,22] The management of oligosymptomatic intracranial lipomas associated with vascular malformation is usually conservative with a satisfactory final outcome and surgical treatment should only be indicated to improve symptoms that cannot be achieved with conservative treatment.[21]

CASE REPORT

A 42 year-old woman with a chronic, periodically appearing headache lasting for the last few years treated successfully with analgesics and with short-term memory problems of unknown duration was referred to the emergency department by radiologist after out-of-hospital brain magnetic resonance imaging (MRI) with suspicion of right middle cerebral artery (MCA) aneurysm and late subacute subarachnoid hemorrhage in the right Sylvian fissure. It is essential to mention that the patient performed this study on her own and no previous clinical examination nor imaging study was conducted. On physical examination, her general condition was surprisingly good in relation to her initial diagnosis and showed moderate severity headache only. The rest of her medical and family history was unremarkable. In the meantime, the radiological consultation of the MRI provided by the patient was carried out. A lesion in the right Sylvian fissure with a dilated right MCA passing across it was visualized. The lesion was non-homogeneously hyperintense in T1W conventional spin echo (T1W CSE), T2W fast spin echo (T2W FSE), and fluid attenuated inversion recovery sequences (FLAIR). Moreover, gradient echo technique susceptibility weighted imaging (SWI) expressed hypointensity and blooming due to susceptibility artifacts. Diffusion-weighted imaging (DWI) did not show diffusion restriction [Figure 1]. Based on the MRI study provided, the possibility of differentiating between adipose tissue and late subacute hemorrhage was inconclusive. Emergency head computed tomography (CT) angiography of the cerebral arteries was performed revealing unruptured fusiform aneurysm of the right MCA located at the border of M1 and M2 segments, extending the length of about 6 mm with maximal radius of the vessel up to 4 mm and with the anatomical variation of the right MCA in the form of trifurcation of the M2 branches was discovered [Figure 2]. Distal half of M1 segment and proximal part of M2 segment of the right MCA were encased by extremely hypodense mass lesion 10 × 13 × 13 mm (−90 to −100 HU) identified as a fat, arranged in Sylvian fissure [Figure 3]. Any signs suggesting blood or macroscopic calcifications were not detected. To definitively confirm the diagnosis of the intracranial lipoma, a control non-contrast MRI was performed with an additional T1W Fat Saturation (T1W FS) sequence, in which there was complete suppression of the fatty tissue [Figure 4]. Taking into account the whole diagnostic pathway, perianeurysmal Sylvian fissure lipoma encasing right MCA trifurcation was diagnosed. Due to good headache control with medications, surgical intervention was abandoned. During hospitalization, the headache subsided, and the patient was discharged home on the 3rd day. After 3 months at the follow-up visit, the patient's condition was uneventful and further follow-up is planned.

DISCUSSION

Intracranial lipomas are rare lesions with the prevalence of 0.1–1.3%.[21,22] Complex nature of the lesion is believed to be closely related to the erroneous differentiation of meninx primitiva into adipose tissue and mature into lipoma. The process takes place at embryonic level and is not precisely understood. As a matter of fact, intracranial lipoma is not neoplasm nor true hamartoma.[25] In contrast to intraventricular tumors such as those found in tuberous sclerosis, intracranial lipomas grow together with the body's general growth. Therefore, symptomatic progression in intracranial lipoma is rarely owing to lesion growth and is much more frequently brought on by regressive changes within surrounding tissues. Its unusual origin and non-neoplastic behavior, respecting and incorporating, rather than displacing, the intracranial vessels and nerves, should make it called congenital malformation.[15,25] Intracranial lipoma is most frequently located in the pericallosal cistern. Other preferential sites of occurrence listed in decreasing the prevalence order include quadrigeminal cistern, suprasellar cistern, and cerebellopontine cistern.[15] Sylvian fissure is among the rarest locations of intracranial lipoma. Truwit and Barkovich reported barely 5% of cases in their microscale study group of 42 patients in 1990, Maiuri *et al*. reported 3.4% out of 203 cases of intracranial lipoma in 1988, and Chao *et al*. revealed just 13 documented cases in the literature review in years 1965–2007.[6,17,25] Several new cases of lipomas were discovered in Sylvian fissure nowadays; however, no big data studies focused on intracranial lipoma placement were performed in the recent years.

Intracranial lipomas are found at any age, mostly incidentally, usually unrelated directly with primary complaint.[8] Size location and proximity to nearby neuronal structures play an

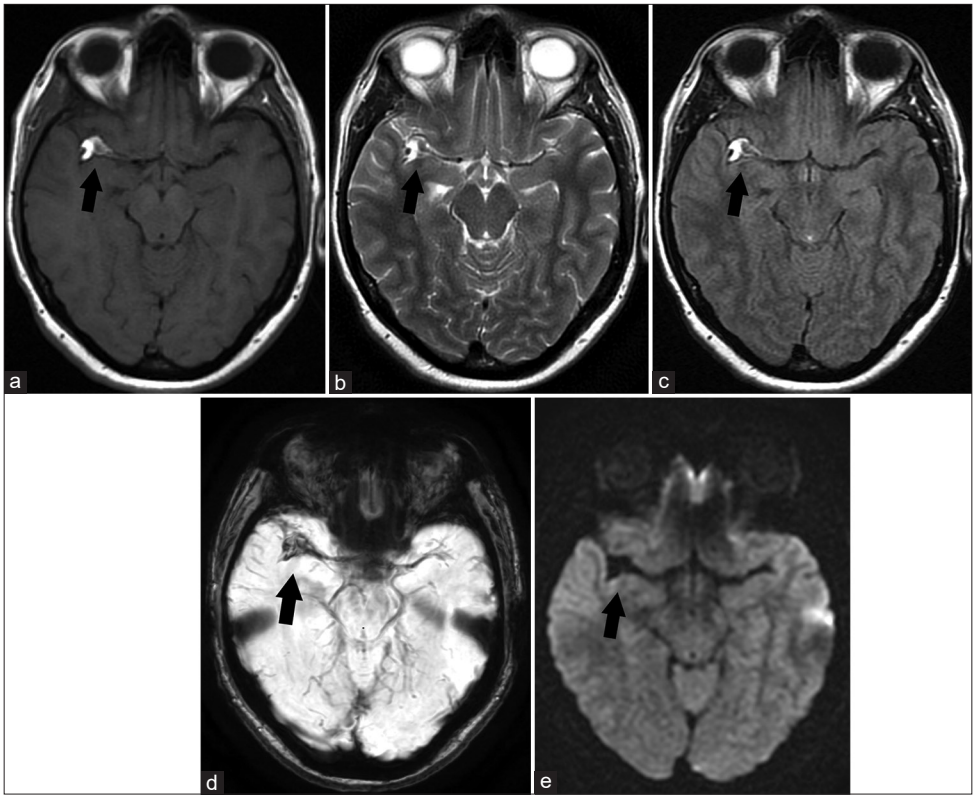

Figure 1: Non-contrast head MRI. Black arrow indicates right Sylvian fissure lipoma encasing right MCA. Axial scans show different signal intensity of the lesion in selected MRI sequences. (a) T1W CSE hyperintense lesion. (b) T2W FSE hyperintense lesion. (c) FLAIR hyperintense lesion. (d) SWI hypointense lesion with blooming artifacts. (e) DWI hypointense lesion. MRI: Magnetic resonance imaging, MCA: Middle cerebral artery, T1W CSE: T1W conventional spin echo, T2W FSE: T2W fast echo spin, SWI: Susceptibility weighted imaging, DWI Diffusion weighed imaging. FLAIR: Fluid-attenuated inversion recovery.

important role in the occurrence of symptoms. In majority, small lipomas located interhemispherically adjacent to the falx cerebri and not associated with dysplasia of the surrounding tissues are usually asymptomatic.[12] Larger lesions and lesions in other intracranial locations might occasionally present symptoms among which persistent headache and epilepsy appear to be the most prevalent.[15,26] Epilepsy is particularly common if the lipoma is located in the Sylvian fissure; Yildiz *et al*. stress this association and emphasize that, in case of seizures, the lipoma in this specific region should be included in the differential diagnosis.[21,27]

Other less common symptoms include vomiting, hemiplegia, vertigo, mental retardation, emotional lability, and cranial nerve impairment.[26] Several cases of lipomas causing obstructive hydrocephalus with symptoms of elevated intracranial pressure have also been reported as a consequence of obstruction of the cerebrospinal fluid flow pathway.[15] The patient presented in our study suffered from moderate in intensity periodical headache and short-term memory problems only. These symptoms are nonspecific, and it is not known whether the intracranial lipoma and vascular malformation were linked with these complaints.

Depending on the source, about 25–60% of the intracranial lipomas are associated with dysgenesis of neuronal brain tissues and vascular malformations.[25,27] Regressive alterations in the form of agenesis and varying degrees of hypogenesis of neural structures and vascular aberrations are closely related to the location of lipomas and therefore most frequently affect the corpus callosum due to the most common placement of lipomas in the pericallosal cistern. Other neural structures such as those of the midbrain, cerebral cortex, or cerebellar vermis can also be affected by dysgenesis.[25] The patient presented in our study had no macroscopic neuronal tissue malformations but was affected by vascular aberration in the form of a fusiform dilatation of the MCA trifurcation. Vascular malformations associated with lipomas are not uncommon, with nerves and vessels being contained within the lesion in 36% of cases.[25] These malformations might include arterial wall defects, profound pathologic vasculature, lateral displacement, tortuous

course, and separation of vessels running parallel to each. Eldevik and Gabrielsen focused on the vascular assessment in association with pericallosal lipoma and found out that fusiform dilatation of pericallosal artery and its branches was present in 19 out of 22 cases.[9] Regarding vascular malformations of the MCA associated with lipoma in the Sylvian fissure, in literature, we found 13 cases, of which 5 cases of saccular aneurysms in the MCA bi/trifurcation, and the case we report of Sylvian fissure lipoma associated with fusiform aneurysm in the MCA with anatomical variation of trifurcation of M2 branches seems to be unique and probably the first one ever reported [Table 1].[1,2,5,6,10,11,13,14,18,20,22,24,27] This finding raises the question of whether a saccular aneurysm of the MCA is more prevalent than a fusiform aneurysm of the MCA when associated with Sylvian fissure lipoma, unlike in case of pericallosal lipoma and fusiform aneurysm of pericallosal artery.

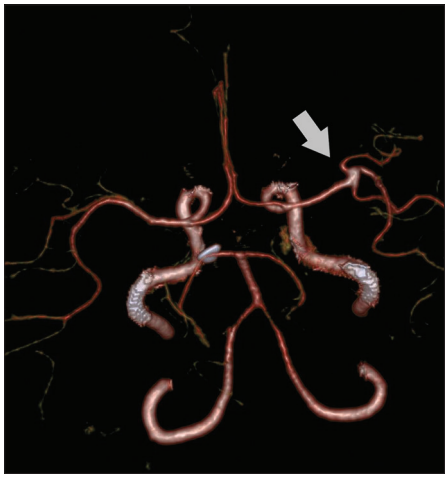

Figure 2: CT angiography in 3D projection of intracranial arteries. The gray arrow indicates the location of fusiform dilatation of the right MCA trifurcation. CT: Computed tomography, MCA: Middle cerebral artery.

Aneurysm formation in association with lipoma does not appear to be incidental; however, the biological mechanism is unclear.[9] Eldevik and Gabrielsen proposed three possible hypotheses. First, aneurysm may arise due to a congenital structural pathology of the blood vessel walls occurring at the same time as the formation of the intracranial lipoma. Another possibility is that the lipoma itself, by its close adhesion to vascular wall might restrict nutritional inflow of cerebrospinal fluid to smooth muscles of cerebral arteries causing their degeneration. Finally, there are suggestions that lipomas might secrete some factors that inhibit arterial smooth muscle growth debilitating arterial wall and by hemodynamic stress result in aneurysm formation.[9] Vascular anatomical variation may be associated with an increase in shear stress which is thought to be a major cause of aneurysmal formation. Although studies in the general population do not show a higher frequency of aneurysm formation in MCA trifurcation compared to MCA bifurcation, the presence of an intracranial lipoma at the MCA trifurcation may possibly have an additional impact on aneurysm occurrence.[4] Further studies should be conducted to confirm this statement.

Intracranial lipomas may mimic late subacute hemorrhage due to similar radiological features on MR-based imaging and this may lead to misdiagnosis. Both these entities present

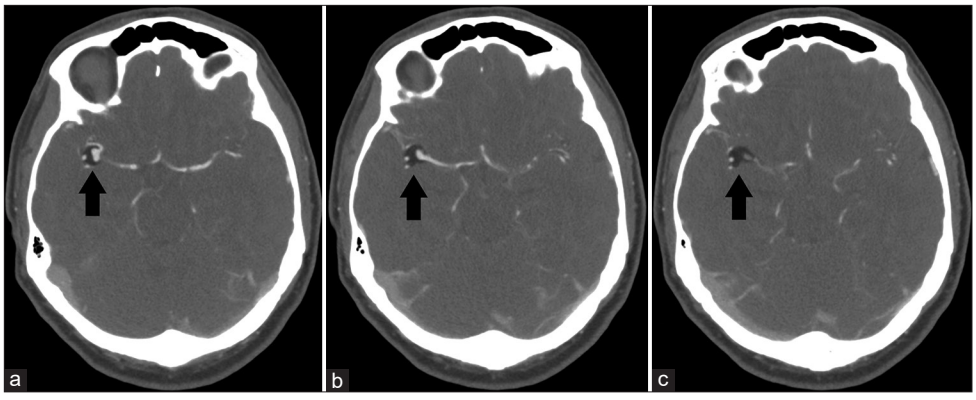

Figure 3: Non-contrast head CT scan in axial projections showing 3 consecutive scans 2,5 mm apart. The black arrow on images (a-c) show a cross-section at different levels of the lesion located in the right Sylvian fissure corresponding to a lipoma and encasing the right MCA. CT: Computed tomography, MCA: Middle cerebral artery.

Figure 4: Axial T1W FS sequence in non-contrast head MRI. The black arrow indicates adipose tissue. MRI: Magnetic resonance imaging.

short T1W and long T2W times expressed as hyperintense signal. Furthermore, in these conditions, gradient echo technique SWI shows hypointensity and blooming artifacts, due to its specific modality in the detection deposits of diamagnetic and paramagnetic substances.[19,22] In case of lipoma and unruptured aneurysm without visible macroscopic calcifications, blooming artifacts of lipoma in the SWI seem to be confusing. Lingegowda *et al*. identify the possible cause of this phenomenon as a contribution of microscopic mineralization and chemical shift at point where fat-water interfaces resulting in signal spatial misregistration.[16]

Bakshi *et al*. came up with the solution of how differentiation between lipoma and late subacute hemorrhage could be achieved.[3] When lipid-based lesions are suspected, authors suggest adding T1W FS sequence to MRI protocol, so it will cause complete signal suppression of adipose tissue, exhibiting marked hypointensity. Another method they propose is to replace the commonly used FSE T2W/PDW with more time consuming CSE T2W/PDW sequences, so the lipomas, unlike in FSE T2W/PDW show hypointense in CSE T2W/PDW. Peter *et al*. also mentioned routinely present in MRI protocols DWI, which can assist in proper distinction. DWI shows strong diffusion restriction in late subacute hemorrhage and loss of signal in lipoma.[19]

On CT, intracranial lipomas appear as non-enhancing mass with very low density ranging between −40 and −100 HU with sporadic calcifications present and no similarities to blood products.[22]

MR of the head is often the diagnostic test of choice for chronic non-traumatic headache.[7] However, this diagnostic imaging in disassociation from the patient's medical history and physical examination can lead to an incorrect initial diagnosis because of the many similarities between various pathologies in MRI. The radiologist's vigilance should be particularly alert to intracranial hemorrhage, especially when a vascular malformation is present. Misdiagnosis in the differentiation of late subacute hemorrhage and intracranial lipoma can be minimized by modifying the standard MR protocol and performing head CT.

Treatment of intracranial lipomas depends on the size and location as well as the presence of symptoms and complications. For the vast majority of intracranial lipomas, the management is based on the simple monitoring of the patient's condition and when symptomatic conservative treatment is recommended. Surgery is limited to very specific conditions, in which conservative treatment is not effective. Attempts at gross total lipoma resection have a relatively high risk of brain injury due to the abundant vascularization of the lesion and the tight adhesion of the lipoma to surrounding nerve structures and vessels.[21] For this reason, the risk-benefit ratio should be considered, and the decision to proceed with surgery should always be individualized.

Symptomatic lipomas of Sylvian fissure associated with vascular malformation of the MCA or its branches are in most cases treated conservatively, and operational approach is rarely applied. The literature describes several cases of different surgical approaches to such complicated intracranial lipomas with generally satisfactory outcomes.[6,10,11] Futami *et al*. performed a right frontotemporal craniotomy and proceeded with a partial resection of the lipoma in the right Sylvian fissure and with clipping of the saccular aneurysm in the right MCA bifurcation. The patient's postoperative outcome was uneventful.[11] Feldman *et al*. described a patient with drug-resistant epilepsy who underwent a modified right pterional craniotomy, and partial resection of a pathologically vascularized lipoma in the right Sylvian fissure was performed. Postoperatively, the patient was seizure free with the aid of anti-epileptic drugs.[10] Chao *et al*. presented a patient with a persistent headache and a lipoma in the right Sylvian fissure. The patient underwent pterion keyhole microsurgery, and gross total resection of the lipoma was performed resulting in resolution of the patient's complaints and uncomplicated postoperative course.[6]

In the presented case, after a complex neurological and neurosurgical consultation, a decision was made to treat the patient conservatively. Closed follow-up with a scheduled angio-RM and angio-CT was ordered to observe the potential progression of the fusiform aneurysm over time. In this case, the risks associated with brain damage far outweighed the benefits of surgery.

Table 1: Sylvian fissure lipoma associated with vascular malformations.

Author, year	Number of individuals	Age/sex	Clinical symptoms	Lipoma location	Type of vascular malformation	Type of treatment	General outcome
Hatashita *et al.*, (1983)[13]	1	20/M	Seizure	Left Sylvian fissure	Stretching of vessels in the region of the mass with no pathological vessels	Surgical partial removal of the lipoma	Improved
Futami *et al.*, (1992)[11]	1	26/F	Headache	Right Sylvian fissure	Saccular aneurysm of the right MCA bifurcation	Surgical partial removal of the lipoma and clipping of the aneurysm	Uneventful
Saatci *et al.*, (2000)[20]	1	17/M	Seizure	Left Sylvian fissure	Abnormal hypervascular network branching out of the left MCA	Conservative with antiepileptic drugs	Improved
Feldman *et al.*, (2001)[10]	1	34/M	Seizure	Right Sylvian fissure	Abnormal hypervascular network branching out of the right MCA	Surgical partial removal of the lipoma	Free of seizures with aid of antiepileptic drugs
Ahmetoğlu *et al.*, (2003)[2]	1	2.5/F	Seizure	Left Sylvian fissure	Abnormal hypervascular network branching out of the left MCA	Conservative with antiepileptic drugs	-
Cerase *et al.*, (2005)[5]	1	27/M	Headache	Right Sylvian fissure	Saccular aneurysm of the right MCA trifurcation	Conservative	Improved
Singh and Shahi, (2005)[18]	1	50/F	Epilepsy	Left Sylvian fissure	Saccular aneurysm of the left MCA bifurcation	Conservative with antiepileptic drugs	Good pharmacological control of epilepsy
Yildiz *et al.*, (2006)[27]	1	55/F	Epilepsy	Right Sylvian fissure	Saccular aneurysm of the right MCA bifurcation	-	-
Chao *et al.*, (2008)[6]	1	57/F	Persistent headache	Right Sylvian fissure	Abnormal hypervascular network branching out of the right MCA	Surgical total removal of the lipoma	Improved
Jabot *et al.*, (2009)[14]	1	44/F	Seizure	Left Sylvian fissure	Left MCA dysplasia	-	-
Schembri, (2013)[22]	1	60/F	Incidental finding (symptoms not related with presence of lipoma or vascular malformation)	Left Sylvian fissure	Saccular aneurysm of the left MCA bifurcation	Conservative	-

(Contd....)

Table 1: *(Continued).*

Author, year	Number of individuals	Age/sex	Clinical symptoms	Lipoma location	Type of vascular malformation	Type of treatment	General outcome
Thakur *et al.*, (2013)[24]	1	25/M	Episodic headache	Right Sylvian fissure	Abnormal hypervascular network branching out of the right MCA	Conservative	Improved
Chandshah *et al.*, (2018)[1]	1	11/M	Seizures	Right Sylvian fissure	Dilatation of the M2 and the M3 segments of the right MCA and angular artery. Posterior branch of the angular artery with focal narrowing	Conservative with antiepileptic drugs	Good pharmacological control of epilepsy
This case (2023)	1	42/F	Headache, short term memory problems	Right Sylvian fissure	Fusiform aneurysm of the right MCA trifurcation	Conservative	Improved

M: Male, F: Female, MCA: Middle cerebral artery

CONCLUSION

The case we present shows that the lipoma can be located in the Sylvian fissure in close proximity to the MCA. The intracranial lipoma can coexist with vascular abnormalities, such as the fusiform aneurysm in the trifurcation of the right MCA. In contrast to lipoma in the pericallosal cistern, lipoma in the Sylvian fissure appears to be more often associated with a saccular aneurysm than with a fusiform aneurysm. Special attention should be given to appropriate modification of the MRI protocol and the performance of a head CT that can establish a definitive diagnosis by ruling out late subacute hemorrhage. The decision on the treatment modality should be made by a comprehensive clinical evaluation of the patient's condition, and surgical treatment should be considered only for symptomatic patients after a prior assessment of its risks and benefits.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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