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Delayed Leukoencephalopathy after Endovascular Embolization of Cerebral Aneurysms

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Abstract: ***Delayed leukoencephalopathy (DL) is a rare and poorly understood complication reported following endovascular embolization of cerebral aneurysms. Although a few isolated case reports exist, the pathomechanism remains unknown. In this case report, we describe the clinical presentation, diagnosis, management, and outcome of a patient who developed DML after undergoing coil embolization for a ruptured cerebral aneurysm, highlighting the importance of early recognition and intervention to improve patient outcomes.***

Keywords: delayed leukoencephalopathy, coil embolization, cerebral aneurysm, endovascular treatment, complication

1. Introduction

Endovascular coil embolization is an established and effective technique for managing cerebral aneurysms, whether unruptured or ruptured. [1] While the procedure is generally considered safe, it is not without potential complications. [2]Periprocedural thromboembolic or hemorrhagic events and delayed aneurysmal rupture have all been documented as possible adverse treatment outcomes. [3] In recent years, delayed leukoencephalopathy (DL) has emerged as a rare and poorly understood complication of coil embolization. [4]

In contrast to the reversible posterior leukoencephalopathy syndrome, DL uniquely involves widespread leukoencephalopathy throughout the hemispheric brain. [5]A limited amount of cases describing leukoencephalopathy have been reported after coil embolization, often attributed to factors such as thrombosis and inflammatory reactions or to underlying conditions like hypertension and subarachnoid haemorrhage. [6]The pathomechanism behind DL remains uncertain, with several proposed etiologies, including granulation reactions caused by foreign body emboli from hydrophilic procedural device coatings, contrast - induced encephalopathy, and sensitivity to materials used in the coils themselves, such as nickel or the bioactive polyglycolic - polylactic acid (PGLA). [7, 8]

Despite increasing reported cases, the overall clinical picture of DL, including its natural history, incidence, onset time, symptoms, treatment, mortality, and morbidity, still needs to be clarified. In this report, we describe a case of DL that developed after coil embolization of an unruptured cerebral aneurysm, highlighting the importance of early recognition and intervention to improve patient outcomes.

2. Case Description

A 38 - year - old male patient with a history of partial coiling for a ruptured right posterior communicating (PcomA) aneurysm and an unruptured right posterior cerebral artery aneurysm presented for a diagnostic follow - up three months after the initial coil embolization. The angiographic assessment revealed recanalization of the embolized aneurysm, prompting the placement of a flow diverter stent to prevent further growth. Two months later, the patient underwent a third procedure to treat the non - ruptured right posterior cerebral aneurysm using a balloon - remodelling technique and coils (Figure 1).

Three months post - procedure, the patient was readmitted due to a one - week history of progressive left mild hemiparesis and right - sided oculomotor nerve palsy. Magnetic resonance imaging (MRI) failed to identify abnormalities correlating with the patient's neurological symptoms (Figure 2). Blood cell count and laboratory serology revealed no related abnormalities.

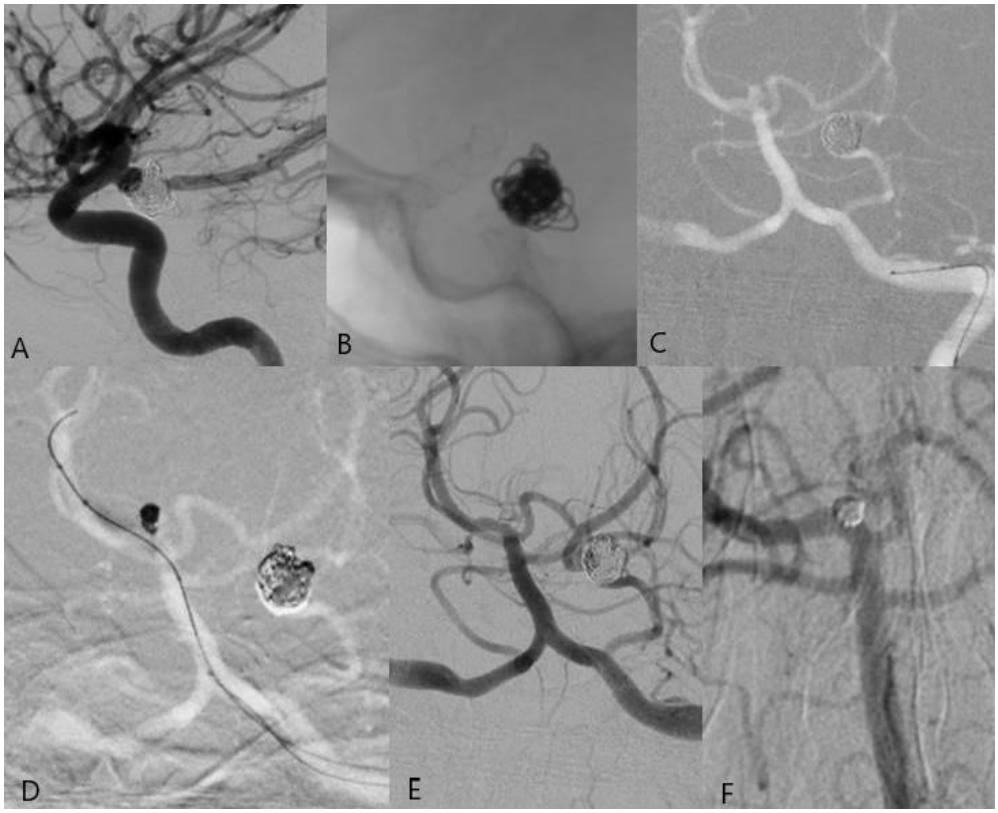

Figure 1: Technical and angiographic aspects of the performed endovascular embolization sessions in a 38 - year - old male patient.

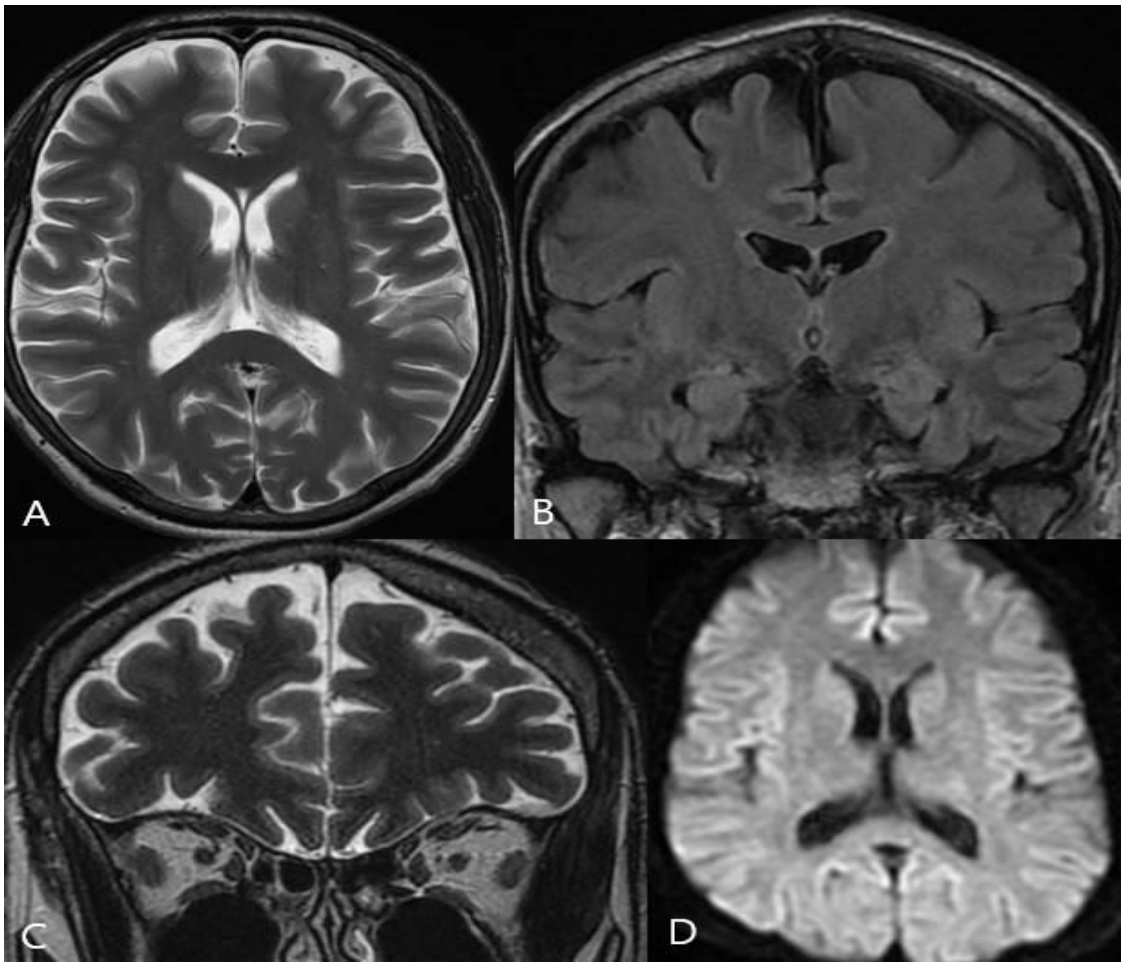

Figure 2: Initial magnetic resonance imaging (MRI) failed to demonstrate any radiological explanation for the mild neurological deficits noted three months after the last embolization session.

The patient was treated with pulse intravenous corticosteroid therapy followed by oral prednisolone, which led to the spontaneous resolution of hemiparesis 11 days after initiating treatment. However, the cranial nerve deficit remained unchanged.

During outpatient follow - up, the patient's oral corticosteroid dosage was gradually reduced every two weeks. However, after a three - month washout period, he was rehospitalized due to progressive cognitive and bilateral visual impairment. MRI revealed diffuse leukoencephalopathy in the left hemisphere, with subcortical high - intensity signals on T2 - weighted, fluid - attenuated inversion recovery, and diffusion - weighted images accompanied by high apparent diffusion coefficient values, suggestive of vasogenic edema. No enhancement by gadolinium was observed on T1 - weighted images (Figure 3).

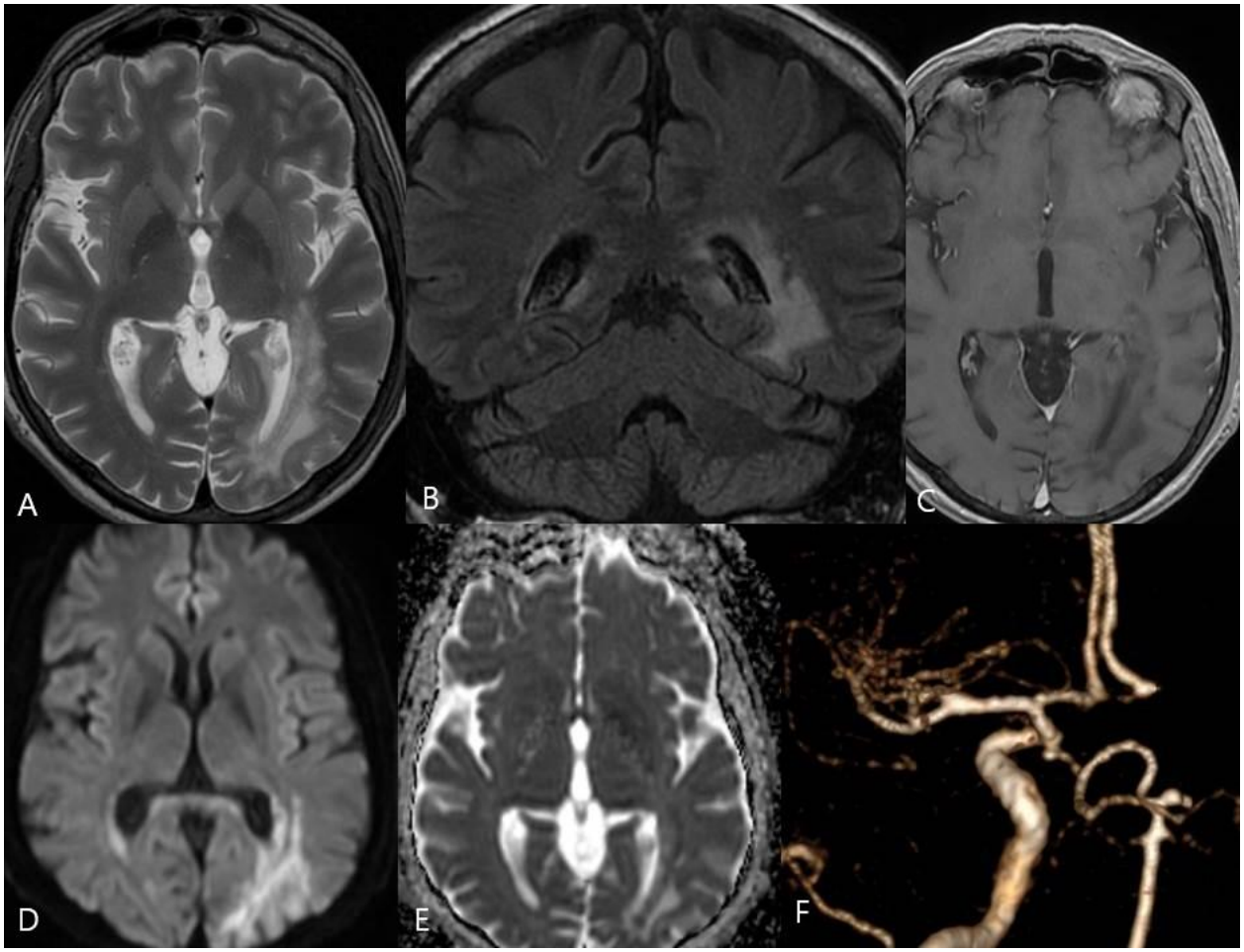

Figure 3: Brain MRI on the rehospitalised day. High - intensity signals on fluid - attenuated inversion recovery (FLAIR) image and diffusion - weighted image with elevated apparent diffusion coefficient in the territory of the left internal carotid artery, indicating vasogenicoedema.

Magnetic resonance arteriography showed no changes in the treated aneurysms, and magnetic resonance venography was normal. No specific autoimmune antibodies were detected, and routine cerebrospinal fluid analyses were unremarkable. Immediate hydrocortisone administration upon readmission did not prevent the progression of visual deficits. However, alternating intravenous methylprednisolone pulse therapy followed by oral prednisolone and glycerol, initiated on the fourth day after rehospitalization, successfully halted the worsening of cognitive symptoms but failed to improve the patient's visual impairment.

Although clinically asymptomatic, a marked deterioration was observed in the follow - up brain MRI. At this stage, the white matter changes were significantly expanded, including T1 spotted and scattered enhancement by gadolinium (figure 4).

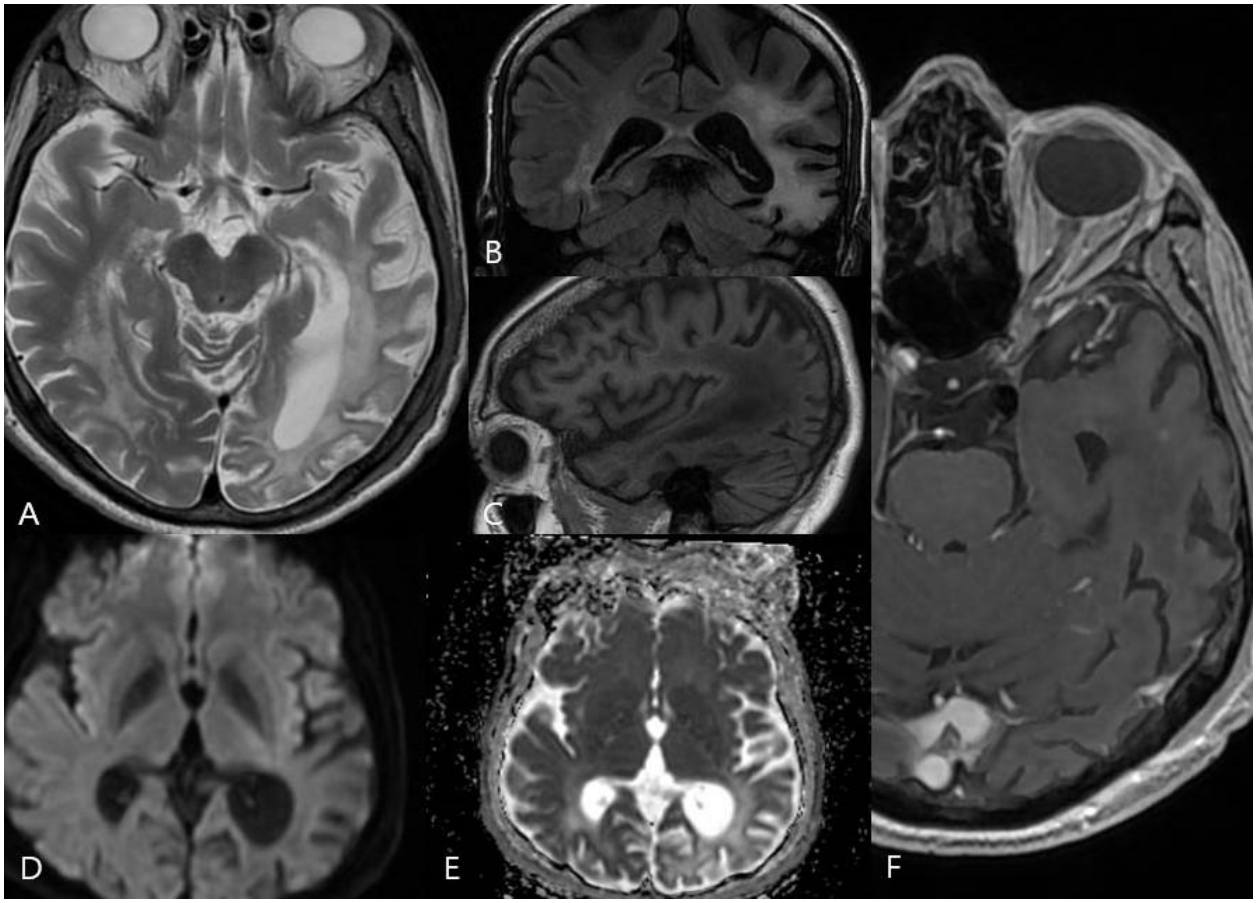

Figure 4: Radiological changes observed in follow - up brain MRI. The images display significant expansion of the white matter changes, including T1 scattered nodular enhancement by gadolinium, reflecting the progression of the leucoencephalopathy.

Subsequently, a cautious reduction and eventual discontinuation of the medical therapy were carried out after 12 months from the onset. The patient's neurological deficits remained unchanged during the last clinical and radiological follow - up.

3. Discussion

Delayed leukoencephalopathy (DL) following endovascular coil embolization of cerebral aneurysms is a rare and poorly understood complication. [9, 10]DL has been described under various names, such as delayed leukoencephalopathy, delayed enhancing lesions, non - ischemic cerebral enhancing and delayed multiple white matter lesions. [11–13]Recently, Ridwan et al. identified 62 cases of DL after endovascular procedures for intracranial aneurysms in the literature. [6]A variety of etiologies have been proposed for this complication, including contrast - induced encephalopathy, granulation reaction caused by foreign body emboli from the hydrophilic coating of procedural devices, and hypersensitivity to coil materials. [4, 14–16]

In our case, the patient presented with progressive neurological symptoms, including left hemiparesis and right - sided oculomotor nerve palsy, three months after the last embolization session. The initial MRI failed to demonstrate any radiological explanation for these deficits, and laboratory investigations did not reveal any significant abnormalities. Treatment with pulse intravenous corticosteroids followed by oral prednisolone led to spontaneous resolution of the hemiparesis, but the cranial nerve deficit remained unchanged.

Upon rehospitalization due to progressive cognitive and bilateral visual impairment, MRI revealed diffuse leukoencephalopathy spreading within the left brain hemisphere. Treatment with corticosteroids could not prevent further progression of the visual deficit, and the patient's neurological symptoms persisted during the last clinical and radiological follow - up. This case highlights the importance of considering DL as a possible complication in patients presenting with neurological deficits following endovascular embolization of cerebral aneurysms.

In some cases of DL, posterior reversible encephalopathy syndrome (PRES) could not be excluded as an etiology for the white matter lesions because their locations were both in the bilateral occipital subcortex, despite the interventions being for basilar apex aneurysms. [5, 17]However, in the present case, the white matter lesion was located only within the ipsilateral hemisphere of the treated aneurysm, displaying a distinctly different pattern from PRES.

The pathogenesis of DL remains unclear, but it is thought to involve a multifactorial process. Some authors have suggested that the development of DL may be related to the use of bioactive coils, which can provoke a more intense inflammatory reaction compared to bare platinum coils. However, in the present case, the coils used were not bioactive, making this explanation less likely. [4]

The treatment of DL is primarily supportive and includes corticosteroids to manage the inflammatory process. In this case, the patient's neurological deficits improved with corticosteroid therapy, but some deficits persisted. This outcome highlights the need for a better understanding of the pathogenesis, early recognition, and optimal management of this rare complication. [6]

As the number of endovascular procedures for cerebral aneurysms continues to increase, it is essential to be aware of potential complications like DL. The present case report adds to the growing body of literature on this rare complication and may help guide future research into its pathogenesis, prevention, and treatment.

In conclusion, DL is a rare complication of endovascular coil embolization for cerebral aneurysms, and its pathogenesis remains poorly understood. The present case highlights the importance of recognizing this complication and considering it as a potential cause of neurological deficits following coil embolization. Further research is needed to better understand the underlying mechanisms and develop strategies for prevention and management of this potentially debilitating condition.

References

[1] Pierot L, Wakhloo AK. Endovascular Treatment of Intracranial Aneurysms: Current Status. *Stroke*.2013; 44 (7): 2046 - 2054. doi: 10.1161/STROKEAHA.113.000733

[2] Ihn YK, Shin SH, Baik SK, Choi IS. Complications of endovascular treatment for intracranial aneurysms: Management and prevention. *Interventional Neuroradiology*.2018; 24 (3): 237 - 245. doi: 10.1177/1591019918758493

[3] Sirakova K, Penkov M, Matanov S, et al. Progressive volume reduction and long - term aneurysmal collapse following flow diversion treatment of giant and symptomatic cerebral aneurysms.: 23.

[4] Ikemura A, Ishibashi T, Otani K, et al. Delayed Leukoencephalopathy: A Rare Complication after Coiling of Cerebral Aneurysms. *AJNR Am J Neuroradiol*.2020; 41 (2): 286 - 292. doi: 10.3174/ajnr. A6386

[5] Fukushima Y, Nakahara I. Delayed leucoencephalopathy after coil embolisation of unruptured cerebral aneurysm. *BMJ Case Reports*. Published online June 23, 2018: bcr - 2018 - 224569. doi: 10.1136/bcr - 2018 - 224569

[6] Ridwan S, Kandyba JA, Schug A, Malsagov E, Karageorgos N, Hans FJ. Delayed Leukoencephalopathy and Foreign Body Reaction After Endovascular Treatment in Patients With Intracranial Aneurysms and Aneurysmal Subarachnoid Hemorrhage—A Systematic Review of the Literature. *Front Surg*.2021; 8: 732603. doi: 10.3389/fsurg.2021.732603

[7] Shotar E, Labeyrie MA, Biondi A, et al. Non - ischemic cerebral enhancing lesions after intracranial aneurysm endovascular repair: a retrospective French national registry. *J NeuroIntervent Surg*.2022; 14 (9): 925 - 930. doi: 10.1136/neurintsurg - 2021 - 017992

[8] Shotar E, Law - Ye B, Baronnet - Chauvet F, et al. Non - ischemic cerebral enhancing lesions secondary to endovascular aneurysm therapy: nickel allergy or foreign body reaction? Case series and review of the literature. *Neuroradiology*.2016; 58 (9): 877 - 885. doi: 10.1007/s00234 - 016 - 1699 - 5

[9] Bakola E, Papagiannopoulou G, Palaiodimou L, et al. Delayed Leucoencephalopathy as a Complication after Endovascular Therapy of Intracranial Aneurysms—A Case Series. *JCM*.2023; 12 (2): 496. doi: 10.3390/jcm12020496

[10] Bakola E, Katsanos AH, Palaiodimou L, et al. Delayed recurrent enhancing white matter lesions complicating coiling of intracranial aneurysm. *Euro J of Neurology*.2021; 28 (7): 2388 - 2391. doi: 10.1111/ene.14844

[11] Sirakov A, Aguilar Pérez M, AlMatter M, Henkes H. Paraophthalmic Internal Carotid Artery Aneurysm: Non - Ischemic Cerebral Enhancing (NICE) Lesions After the Endovascular Treatment of an Incidental Paraophthalmic Aneurysm with Flow Diverters and Coils; Conservative Management, with Resolution of the Pathological Cerebral Findings and Clinical Recovery During Mid - Term Follow - Up. In: Henkes H, Lylyk P, Ganslandt O, eds. *The Aneurysm Casebook*. Springer International Publishing; 2020: 1 - 12. doi: 10.1007/978 - 3 - 319 - 70267 - 4_142 - 1

[12] Cruz JP, Marotta T, O'Kelly C, et al. Enhancing Brain Lesions after Endovascular Treatment of Aneurysms. *AJNR Am J Neuroradiol*.2014; 35 (10): 1954 - 1958. doi: 10.3174/ajnr. A3976

[13] Bayas A, Christ M, Berlis A, et al. Incidence, clinical spectrum, and immunotherapy of non - ischemic cerebral enhancing lesions after endovascular therapy. *Ther Adv Neurol Disord*.2022; 15: 175628642110723. doi: 10.1177/17562864211072372

[14] Lobotesis K, Mahady K, Ganesalingam J, et al. Coiling - associated delayed cerebral hypersensitivity: Is nickel the link? *Neurology*.2015; 84 (1): 97 - 99. doi: 10.1212/WNL.0000000000001106

[15] Shapiro M, Ollenschleger MD, Baccin C, et al. Foreign Body Emboli following Cerebrovascular Interventions: Clinical, Radiographic, and Histopathologic Features. *AJNR Am J Neuroradiol*.2015; 36 (11): 2121 - 2126. doi: 10.3174/ajnr. A4415

[16] Mellemkjær T, Chandra RV, Speiser L, Ulhøi BP, Simonsen CZ. Delayed leukoencephalopathy from suspected polymer embolism after neuroendovascular procedures. *Neuroradiol J*.2021; 34 (4): 373 - 378. doi: 10.1177/19714009211029172

[17] Naito H, Yamazaki Y, Takahashi T, Ochi K, Kiura Y, Matsumoto M. A case of posterior reversible encephalopathy syndrome associated with coil embolization of a basilar apex aneurysm. *Rinsho Shinkeigaku*.2013; 53 (7): 518 - 525. doi: 10.5692/clinicalneurol.53.518