**Scholars Journal of Medical Case Reports**
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

**Neurology**

# An Unusual Presentation of Osmotic Myelinolysis Syndrome in a Type 1 Diabetes Patient Involving the Spinal Cord

Malika Berrada[1*], Nour El Houda Sguiar El Hamadani[1], Hicham El Otmani[1, 2], Mohammed Abdoh Rafai[1, 3], Bouchra El Moutawakil[1, 2]

[1]Département De Neurologie, Hopital Ibn Rochd, Université Hassan II, Faculté de Médecine et de Pharmacie- Rue Tarik Ibn Ziad, 20360, Casablanca, Marocco
[2]Laboratoire De Génétique et Pathologie Moléculaire, Université Hassan II, Faculté de Médecine et de Pharmacie- Rue Tarik Ibn Ziad, 20360, Casablanca, Marocco
[3]Laboratoire De Recherche Sur Les Maladies Du Système Nerveux Neurosensorielles et handicap, Université Hassan II, Faculté de Médecine et de Pharmacie- Rue Tarik Ibn Ziad, 20360, Casablanca, Marocco

**DOI:** 10.36347/sjmcr.2022.v10i03.003 | **Received:** 17.01.2022 | **Accepted:** 28.02.2022 | **Published:** 04.03.2022

***Corresponding author:** Malika Berrada
Département De Neurologie, Hopital Ibn Rochd, Université Hassan II, Faculté de Médecine et de Pharmacie- Rue Tarik Ibn Ziad, 20360, Casablanca, Marocco

**Abstract** **Case Report**

Osmotic myelinolysis (OM) affecting the central nervous system typically occurs in the pons with or without involvement of other brain regions. Extrapontine myelinolysis can rarely occur in the spinal cord. Diabetic ketoacidosis is an uncommon and scarcely cause of OM. We report a case of a patient with type 1 diabetes who presented with an unusual presentation of OM syndrome affecting the pons and spinal cord and describe the clinical and radiographic findings in the context of other scarce reported cases.

**Keywords:** Diabetes, myelinolysis, spinal, magnetic resonance imaging.



## INTRODUCTION

Osmotic myelinolysis (OM) affecting the central nervous system typically occurs in the pons (CPM) with or without involvement of other brain regions. Extrapontine myelinolysis (EPM) can rarely occur in the spinal cord. We report a case of young man with CPM and EPM affecting the spinal cord in an uncommun context of ketoacidosis and describe the clinical and radiographic findings in the context of other reported cases.

## CASE REPORT

We report the case of a 32-year-old man with a history of long-standing poorly controlled type 1 diabetes who was brought to the emergency room for diabetic ketoacidosis (DKA). His family stated that he was hospitalized two months before for a previous (DKA) (570 mg/dl blood glucose) corrected within 24 hours to130mg/dl and he kept as sequelae an unsteady gait, dysarthria, dysphagia and sphincteric disorders with no tendency to progression. Blood investigations at admission revealed hyperglycemia 580mg/dl (70-99), sodium 158 mmol/L, potassium 4.5 mmol/L, HBA1c at 12% (6,5-7,5) and blood gas sample with pH 7.25 (7,35-7,45) .The patient received intraveinous rehydration with insuline therapy, monitoring of blood electrolytes was performed until his (DKA) was completely corrected. After the life-threatening state was averted and general condition improved, patient was transferred to our neurological department for investigation. On examination, patient was alert with spastic tetraparesis, gait ataxia and pseudobulbar symptoms. Brain MRI showed symmetrical T2/FLAIR weighted hyperintensities and in diffusion weighted imaging sequences centered in the pons, upper mild brain and superior cerebellar peduncle and hypointensities in T1 sequences (Figure 1a & 1b). MRI of the cervical spine showed multiple non-contiguous spinal cord lesions from the cervico-medullary junction to the level of the fifth cervical vertebra (Figure 1c, & 1d). The differential diagnoses of these radiologic findings include MOGAD and NMOSD, their serum antibody titers were negative. Clinical and radiological presentation was consistent with CPM and EPM. The neurological status remained stable.

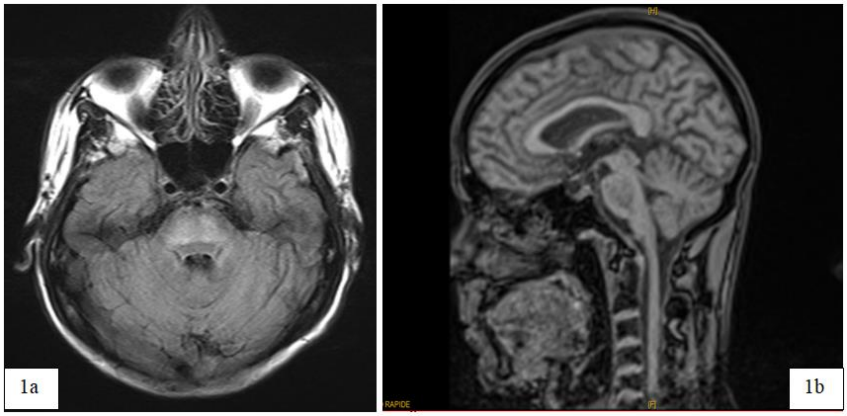


**Figure 1: Brain Magnetic resonance imaging (MRI) showed symmetrical, central hyperintensities in fluid attenuated inversion recovery (FLAIR/ axial) (1a) and hypointensities in T1 /Sagittal (1b) in the pons and upper mild brain without mass effect**

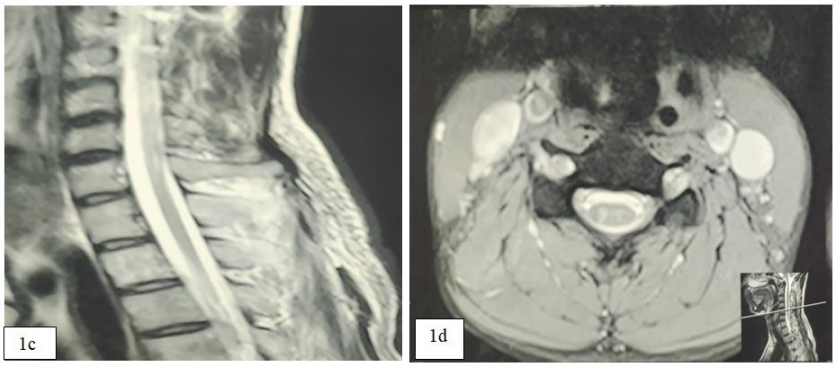


**Figure 1: (c sagittal/d: axial) MRI of the cervical spine showed multiple non-contiguous spinal cord expansive lesions from the cervico-medullary junction to the level of the fifth cervical vertebra**

## DISCUSSION

Osmotic myelinolysis (OM) affecting the central nervous system (CNS) typically occurs in the pons (CPM) with or without involvement of other brain regions. Extrapontine myelinolysis (EPM) can rarely occur in the spinal cord. The underlying pathology of OM is characterized by rapid changes in osmotic pressure wich presumably induce apoptosis of astrocytes and disrupt the blood-brain barrier. As result, cytotoxic factors in blood become able to enter the brain, injuring oligodendrocytes and leading to demyelination [1]. A variety of potential etiologies have been identified (alcoholism, malnutrition…) [2]. DKA is an uncommon cause of OM with uncertain physiopathology [3]. Here, it is plausible that a rapid drop in osmolality in a chronic state of high osmolality (uncontrolled diabetes) led to CPM and EPM. Myelinolysis is best appreciated on MRI, DWI sequences are extremely useful for early diagnosing CPM [1]. MRI findings in OM usually shows symmetric signal hyperintensity in the central pons on T2/FLAIR weighted imaging and hypointensity signal in T1 imaging [4]. In our literature review using pubmed database, we found only 4 publications encompassing 4 cases of spinal cord involvement in OM [2, 4, 5]. Cervical segment of the spinal cord was involved in all the cases. On MRI imaging, T2 hyperintensity was contiguous in 3 cases and patchy in 1 case. CPM associated with EPM was present in 3 cases and brain MRI was normal in the other case. Our case report clinical and radiological presentation was consistent with the literature.

## CONCLUSION

EPM involving the spinal cord in a context of DKA is rare, a slower correction of hyperglycemia possibly could have prevented it.

**Declarations:**
**Conflict of interest:** None

**Acknowledgments:** We wish to thank the patient and his family for their permission to publish this manuscript.
Funding: Not applicable.

## REFERENCES

1. Kusumoto, K., Koriyama, N., Kojima, N., Ikeda, M., & Nishio, Y. (2020). Central pontine myelinolysis during treatment of hyperglycemic hyperosmolar syndrome: a case report. *Clinical Diabetes and Endocrinology*, *6*(1), 1-6.
2. Biller, J., Hornik, A., Rodriguez-Porcel, F., Agha, C., Flaster, M., Morales Vidal, S. G., ... & Lee, J. (2012). Central and extrapontine myelinolysis affecting the brain and spinal cord. An unusual

presentation of pancreatic encephalopathy. *Frontiers in neurology*, *3*, 135.

3. Caldito, N. G., Karim, N., & Gebreyohanns, M. (2021). Teaching NeuroImages: Central Pontine Myelinolysis in Diabetic Ketoacidosis. *Neurology*, 97(19), e1971-e1972.
4. Jacob, S., Gupta, H., Nikolic, D., Gundogdu, B., & Ong, S. (2014). Central pontine and extrapontine myelinolysis: the Great Masquerader—an autopsy case report. *Case reports in neurological medicine*, *2014*, 745347.
5. Rodriguez, L., & Silliman, S. (2014). Longitudinally Extensive Spinal Cord Osmotic Myelinolysis (P1. 038). Neurology, 82(10 Supplement) P1.038.