

Long Term Sustaining Enhanced Infundibulum-hypophysitis: A Case Report

Yasushi Shibata¹, MD, PhD

¹ Department of Neurosurgery, Mito Medical Center, University of Tsukuba, Mito, Ibaraki, 3100015, Japan

Conflict-of-interest statement: The author(s) declare(s) that there is no conflict of interest regarding the publication of this paper.

Open-Access: This article is an open-access article which was selected by an in-house editor and fully peer-reviewed by external reviewers. It is distributed in accordance with the Creative Commons Attribution Non Commercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited and the use is non-commercial. See: <http://creativecommons.org/licenses/by-nc/4.0/>

Correspondence to: Yasushi Shibata, Department of Neurosurgery, Mito Medical Center, University of Tsukuba, Mito, Ibaraki, 3100015, Japan.
Email: yshibata@md.tsukuba.ac.jp

Received: August 1, 2021

Revised: September 16, 2021

Accepted: September 18, 2021

Published online: September 26, 2021

ABSTRACT

INTRODUCTION: Hypophysitis is a non-infectious chronic inflammation. Lymphocytic hypophysitis which is the most frequent adult hypophysitis naturally remit within 2 years.

CASE REPORT: The patient is female. She has had a bilateral occasional headache since her twenties. Her polyuria and thirsty started at 32 years old. She was diagnosed with central diabetes insipidus and desmopressin was started at 33 years old. Magnetic resonance imaging (MRI) showed swelling of the pituitary stalk and 3rd ventricle floor, which were homogeneously enhanced. Brain MRI showed stable disease until 9 years after symptom onset. At 10 years since symptom onset, the decrease of the enhanced lesion was observed.

DISCUSSION: Because of self-limiting benign natural history, the biopsy is generally not applied for the patients under the diagnosis of lymphocytic hypophysitis. Our case showed the problem of the watch and scan strategy.

CONCLUSION: We reported the case of infundibulum-hypophysitis those enhanced lesions sustained for 9 years. This case report demonstrated the problem of the watch and scan strategy.

Key words: Hypophysitis; Enhance; MRI

© 2021 The Author(s). Published by ACT Publishing Group Ltd. All rights reserved.

Shibata Y. Long Term Sustaining Enhanced Infundibulum-hypophysitis: A Case Report. *International Journal of Radiology* 2021; **8(1)**: 326-327 Available from: URL: <http://www.ghrnet.org/index.php/ijr/article/view/3212>

CASE REPORT

Hypophysitis is non-infectious chronic inflammation. Lymphocytic hypophysitis which is the most frequent adult hypophysitis naturally remit within 2 years^[1]. We experienced the case of infundibulum-hypophysitis those enhanced lesions sustained for 9 years.

The patient is female. She has had a bilateral occasional headache since her twenties. She does not have migraine symptoms such as nausea and photo-hypersensitivity. Her polyuria and thirsty started at 32 years old. She was diagnosed with central diabetes insipidus and desmopressin was started at 33 years old. She had no history of pregnancy. She was also diagnosed and treated with pan-hypopituitarism, diabetes mellitus, obesity, dyslipidemia. She had an irregular menstruation cycle. Magnetic resonance imaging (MRI) showed swelling of the pituitary stalk and 3rd ventricle floor, which were homogeneously enhanced (Figure 1A). Optic chiasm compression was not observed and she did not show visual symptoms. Her history and physical examination did not show malignancy, infection, auto-immune disease, and granulomatous diseases. To differentiate from germinoma and IgG4 related diseases, we checked IgG, IgG4, AFP (alpha fetoprotein) in venous blood, and β -hCG (β -human chorionic gonadotropin) in urine, these were all normal. She did not have a history of the administration of an immune-checkpoint inhibitor. We diagnosed her with lymphocytic infundibulum-hypophysitis and planned a watch and scan strategy without biopsy and medication such as a steroid. Brain MRI showed stable disease until 9 years after symptom onset (Figure 1B-D). At 10 years since symptom onset, the decrease of the enhanced lesion was

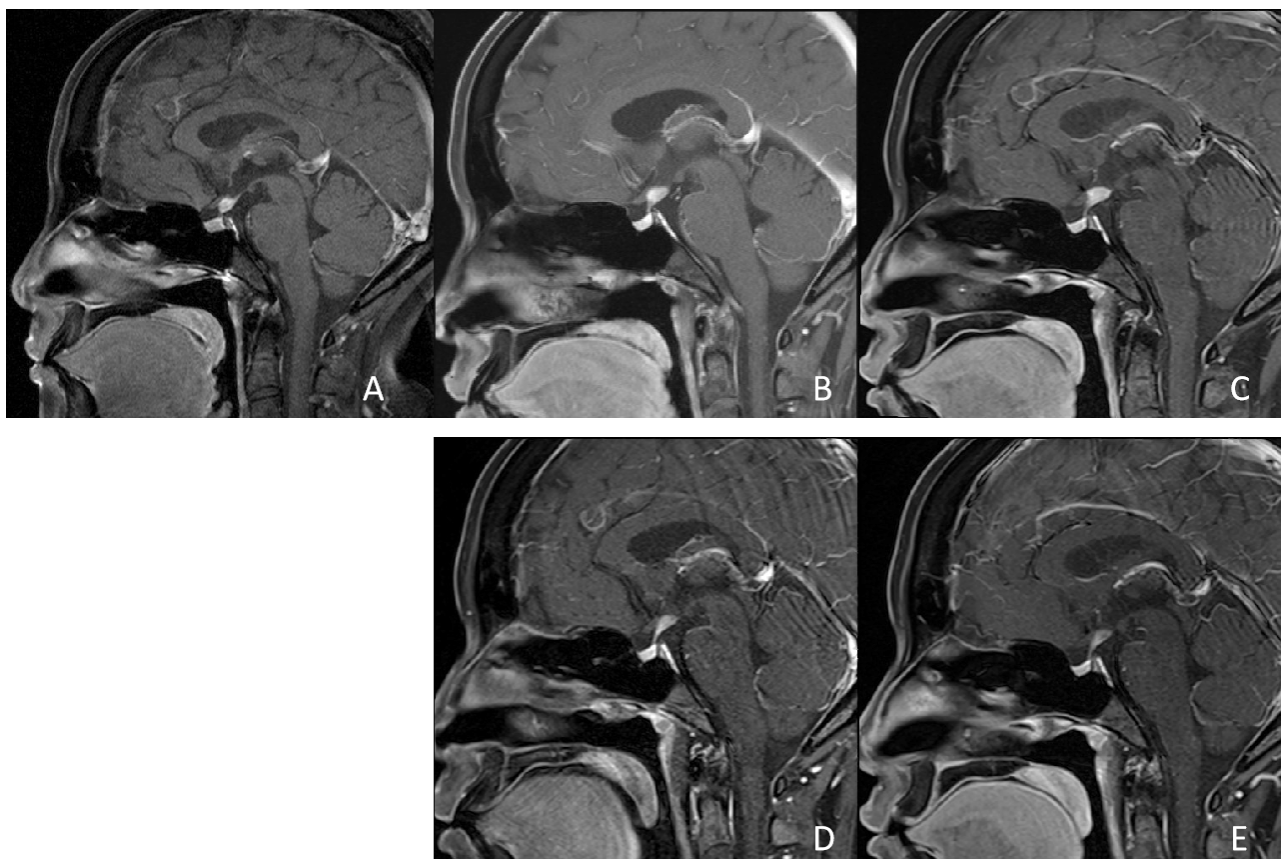


Figure 1 Gadolinium enhanced T1 weighted sagittal MRI at 34(A), 37(B), 41(C), 43(D), 44(E) year-old.

observed (Figure 1E). Her symptoms including diabetes insipidus and headache were not significantly changed. She suffered from infertility, however, she delivered a baby after in-vitro fertilization at 42 years old. The patient has provided informed consent for publication of the case.

DISCUSSION

The differential diagnosis of enhanced pituitary lesions includes tumor, infection, granulomatous and auto-immune diseases^[1]. Systemic and non-invasive examinations sometimes do not bring a clear diagnosis. Lymphocytic hypophysitis is the most frequent adult hypophysitis. Because of self-limiting benign natural history, the biopsy is generally not applied for the patients under the diagnosis of lymphocytic hypophysitis. Our case showed the problem of the watch and scan strategy.

Iwama et al. reported a biomarker for the diagnosis of lymphocytic hypophysitis^[2]. At present, this antibody is not commercially available. The development and commercial supply of a diagnostic biomarker for lymphocytic hypophysitis is expected.

CONCLUSION

We reported the case of infundibulum-hypophysitis those enhanced lesions sustained for 9 years. This case report demonstrated the problem of the watch and scan strategy.

REFERENCES

1. Joshi MN, Whitelaw BC, Carroll PV. MECHANISMS IN ENDOCRINOLOGY: Hypophysitis: diagnosis and treatment. *European Journal of Endocrinology*. 01 Sep. 2018 2018; **179**(3): R151. [PMID: 29880706]; [DOI: 10.1530/EJE-17-0009]
2. Iwama S, Sugimura Y, Kiyota A, Kato T, Enomoto A, Suzuki H, Iwata N, Takeuchi S, Nakashima K, Takagi H, Izumida H, Ochiai H, Fujisawa H, Suga H, Arima H, Shimoyama Y, Takahashi M, Nishioka H, Ishikawa S, Shimatsu A, Caturegli P, Oiso Y. Rabphilin-3A as a Targeted Autoantigen in Lymphocytic Infundibulo-neurohypophysitis. *The Journal of Clinical Endocrinology & Metabolism*. 2015; 100(7): E946-E954. [PMID: 25919460]; [PMCID: PMC5393526]; [DOI: 10.1210/jc.2014-4209]