

Case Report

Sheehan's syndrome of a 50 years old female with bipolar disorder: a case report

Ankit Anand¹, Rajveer Singh², Abhishek Kumar Gupta^{2*}, Ajay Kumar², Hritika², Priyanka Kumari², Shiwani Shukla², Yuvika Kataria¹

¹Department of Medicine, JNUIMSRC, Jaipur, Rajasthan, India

²School of Pharmaceutical Sciences, Jaipur National University Jaipur, Rajasthan, India

Received: 18 February 2024

Accepted: 12 March 2024

*Correspondence:

Abhishek Kumar Gupta,

E-mail: kumar.abhishek199800@gmail.com

Copyright: © the author(s), publisher and licensee Medip Academy. This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

ABSTRACT

Empty Sella syndrome (ESS) pertains to a phenomenon in which the Turkish saddle seems through radiography to be larger and perhaps partially or entirely filled with brain fluids. It can be a major illness/ develop as a result of pituitary medical procedures, radiation therapy cerebral infarction/bleeding process. A 50 years old female patient was admitted in emergency medicine ward with chief complaint of pain in abdomen, weakness and vomiting. MRI scan of pituitary cerebrospinal fluid (CSF) signal intensity is seen in Sella with no visible anterior pituitary gland tissue noted-likely suggestive of secondary empty Sella (SES). Long-term steroid use is principal therapy for adrenocorticotrophic hormone deficiency (IAD). In this instance, ESS, which can be primary/secondary, also noticed.

Keywords: CSF, ESS, Cortisol, Secondary ESS, Isolated adrenocorticotrophic hormone

INTRODUCTION

The expression "empty Sella syndrome" (ESS) pertains to a phenomenon in which the Turkish saddle seems through radiography to be larger and perhaps partially or entirely filled with brain fluids.¹ The most typical sign of ESS is a headache. The headache is typically deep, dull, and centred. Prior to imaging scan, headache can occasionally be extremely intense and be accompanied by giddiness and vomiting.² The brain's pituitary gland is following this compressed and displaced as a result. It can be a major illness/develop as result of pituitary medical procedures, radiation therapy cerebral infarction, or the bleeding process.³ In year 1951, Busch identified for first time a neurological disorder commonly referred to as ESS, where empty CSF-filled Sella turcica generates ordinarily symmetrical structure of pituitary gland to fluctuate/ become flattened.⁴ In autopsy studies, percentage of people with symptoms of Empty Sella ranges from 6-20%; in clinical practise, it is also frequently identified, being reported in 8 to 35% of general population. An

increase in incidence over past few decades may be attributable to advancements in and availability of neuroimaging techniques (up to 38% in MRI studies).⁵⁻⁸ In present study, we discuss a case of SES in 50-year-old female patient with pituitary stalk duplication. According to its definition, PES is herniation of subarachnoid space into Sella turcica as a result of a CSF signal intensity is seen in Sella with no visible anterior pituitary gland tissue noted empty Sella. In primary empty Sella are most likely to experience hypo-pituitarism, with 50% frequency. ESS is also usually accompanied by hyperprolactinemia because hypothalamus area does not produce enough dopamine to function as an inhibitor.

CASE REPORT

A 50 years old female patient was admitted in the emergency medicine ward with chief complaint of pain in abdomen, weakness and vomiting since 10 days. History of present illness was pain in abdomen and vomiting was gradually progressive in nature. Contributory past history

of hypertension and depression and there was history of left cradling bias (LCB) 22 years back, patient had menses only once after LCB, child died at birth and there was history of bipolar affective disorder with 1st episode at 20 years back followed by 2nd episode at 8 years back but the treatment was not taken at that time but got admitted in hospital 4 years back when there was history of 1 more episode and patient got treated with olanzapine 5 mg and tablet clonazepam 0.5 mg and now patient is off from these treatment from last 3.5 years. Surgery named laparoscopic cholecystectomy was performed 10 days ago before hospital readmission. Laboratory investigation as of serum cortisol level was found to be reduced (28.4 ng/ml) and as per report of thyroid profile free T3 and T4 was found quite low as 1.53 pg/ml and 0.610 ng/dl respectively and as per biochemistry report hyponatremia was noted (Na^+ :-121.0 mmol/L and according to blood sugar patient was recurrent hypoglycemic hematology report shows Hb (11.2 gm/dl), TLC (15.6 thousand/cumm), neutrophils (91.1%), lymphocytes (4.4%). In radiological investigation as per MRI scan of pituitary CSF signal intensity is seen in Sella with no visible anterior pituitary gland tissue noted-likely suggestive of SES. Based on history examination and investigation, diagnosis of secondary ESS was made Figure 1 and 2. Patient was treated with drug hydrocortisone 100 mg TDS titrated by dose of 50 mg BD followed by 40 mg to 20 mg and levothyroxine 50 mcg OD was given under expert guidance of consultant.

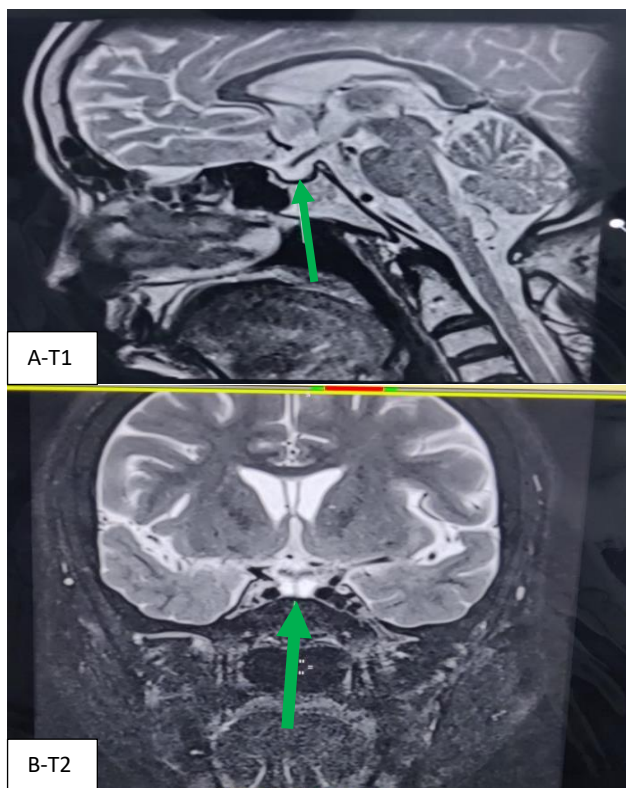


Figure 1 (A and B): An axial view of a T1-weighted magnetic resonance scan of brain. A green arrow denotes pituitary's flattened position. T2-Coronal view region-CSF filling empty Sella.

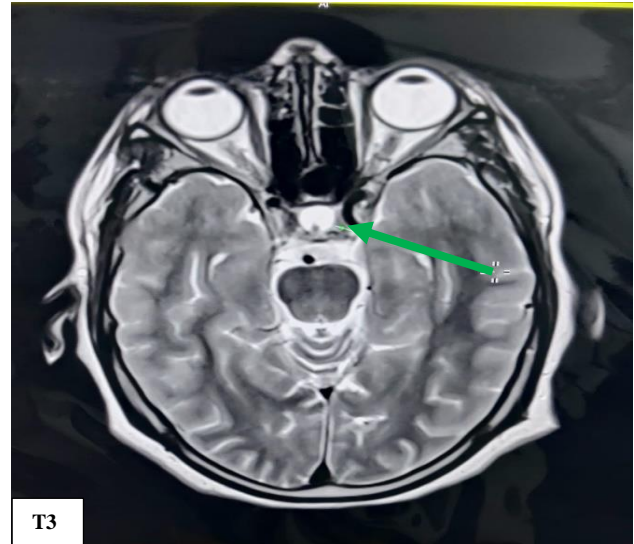


Figure 2: T3-cronal T3-Weighted MRI image green arrow located a presence of empty Sella.

DISCUSSION

In this instance, secondary adrenocortical insufficiency brought on by immune checkpoint inhibitor (ICI) related Isolated IAD was identified, and the patient's prognosis was enhanced by steroid therapy.⁹ Patients with primary empty Sella are most likely to experience hypopituitarism, with a 50% frequency. ESS is also usually accompanied by hyperprolactinemia because the hypothalamus area does not produce enough dopamine to function as an inhibitor. Blood pressure must be maintained by pituitary hormones from both the anterior and posterior lobes, especially during times of stress. Notably, the production of thyroxine and cortisol, both of which have permissive effects on the activities of catecholamines, depends on thyroid stimulating hormone (TSH) and ACTH from the anterior lobe. In a rare instance, Lamont and colleagues described a patient with ESS following cardiac surgery who had lowered levels of both ACTH and vasopressin. Following surgery, their patient had hypotension, which was treated with a cortisol and vasopressin supplement. Panhypopituitarism and ESS are seldom observed together. With an impacted anterior lobe, a normal TSH and free triiodothyronine (FT3), a mildly elevated prolactin, a low normal ACTH, and a low cortisol level, our patient also had an affected anterior lobe.^{6,10} The pathogenic conditions that cause SES, on the other hand, include genetic diseases, hypophysis, pituitary stroke, Sheehan's syndrome, pituitary surgery or radiation therapy, brain damage, and infection.¹¹ The long-term steroid use is the principal therapy for IAD. In this instance, ESS, which can be primary or secondary, was also noticed. In addition to radiation, medications, and surgery, secondary ESS in the pituitary can result from spontaneous necrosis, infectious, autoimmune, or traumatic causes.¹² With no anterior pituitary gland tissue visible, the Sella shows a high CSF signal intensity during radiological examination, which is

likely an indication of a SES. The patient was 50 years old female admitted in the emergency medicine ward with the chief complaint of pain in abdomen, weakness and vomiting since 10 days. In radiological investigation as per MRI scan of pituitary CSF signal intensity is seen in Sella with no visible anterior pituitary gland tissue noted-likely suggestive of SES based on the history examination and investigation, diagnosis of secondary ESS was made. The patient was treated with the drug hydrocortisone 100 mg TDS titrated by dose of 50 mg BD followed by 40 mg to 20 mg and levothyroxine 50 mcg OD was given under the expert guidance of consultant. The patient received care from a team of healthcare professionals including clinical pharmacists, nursing staff, and physician. After getting proper care from the health care professionals her symptoms got relieved and she must take medicines as prescribed. She was counseled properly to take care and follow the instructions of health care professionals.

CONCLUSION

In this report, we discussed a rare case of ESS. Our study reports that her symptoms got relieved after taking treatment and proper medication as suggested by the physician. This case highlights the importance of steroids use and early diagnosis as in detecting the condition and it must be noted for improving outcomes of steroids treatment. Early diagnosis and treatment is indicated to prevent any further complication.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

REFERENCES

1. Agrawal JK, Sahay RK, Bhadada SK, Reddy VS, Agrawal NK. Empty Sella syndrome. Indian Academy Clin Med. 2001;2(3):198-02.
2. Sethuraman VK, Viswanathan S, Aghoram R. Refractory hypoglycemia and seizures as the initial presenting manifestation of empty Sella syndrome. Cureus. 2018;10:e2803.
3. Kumar K, Khalid M, Fadhil A, Lamba P, Basha S, Ibrahim S. Empty Sella Syndrome: A Case Report and Literature Review. Neurosci Med. 2015;6:42-5.
4. Sachdev Y, Evered DC, Appleby A, Hall R. The empty Sella syndrome. Postgrad Med J. 1976;52(613):703-5.
5. Foresti M, Guidali A, Susanna P. Primary Empty Sella. Incidence in 500 Asymptomatic Subjects Examined with Magnetic Resonance. La Radiol Med. 1991;81(6):803-7.
6. Auer MK, Stieg MR, Crispin A, Sievers C, Stalla GK, Kopczak A. Primary Empty Sella Syndrome and the Prevalence of Hormonal Dysregulation. Dtsch Arztebl Int. 2018;115(7):99-105.
7. Chiloiro S, Giampietro A, Bianchi A, Tartaglione T, Capobianco A, Anile C, et al. Diagnosis of Endocrine Disease: Primary Empty Sella: A Comprehensive Review. Eur J Endocrinol. 2017;177(6):R275-85.
8. Sage MR, Blumbergs PC. Primary Empty Sella Turcica: A Radiological Anatomical Correlation. Australas Radiol. 2000;44(3):341-8.
9. Percik R, Shlomai G, Tirosh A, Amit T, Raya L-A, Yael E, et al. Isolated autoimmune adrenocorticotrophic hormone deficiency: from a rare disease to the dominant cause of adrenal insufficiency related to check point inhibitors. Autoimmun Rev. 2020;19(2):102454.
10. Lamont SN, McBride WT, Bill KM, Varadarajan B. Profound vasodilatory hypotension in a patient with known empty Sella syndrome following cardiac surgery. Anaesthesia. 2007;62(8):846-9.
11. Carosi G, Brunetti A, Mangone A, Baldelli R, Tresoldi A, Del Sindaco G, et al. A Multicenter Cohort Study in Patients with Primary Empty Sella: Hormonal and Neuroradiological Features Over a Long Follow-Up. Front Endocrinol (Lausanne). 2022;13:925378.
12. De Marinis L, Bonadonna S, Bianchi A, Maira G, Giustina A. Primary empty Sella. J Clin Endocrinol Metab. 2005;90(9):5471-7.

Cite this article as: Anand A, Singh R, Gupta AK, Kumar A, Hritika, Kumari P, et al. Sheehan's syndrome of a 50 years old female with bipolar disorder: a case report. Int J Sci Rep 2024;10(5):173-5.