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Case Report

An unusual case of hypopituitarism

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ABSTRACT

Lymphocytic hypophysitis represents a difficult diagnostic and therapeutic challenge. The clinical presentation of this inflammatory condition may mimic that of a pituitary adenoma with apoplexy. We report a case of a 45 years old male patient with acute adrenal crisis complicating chronic hypopituitarism secondary to lymphocytic hypophysitis. The patient was managed acutely with high dose steroids and supportive measures, and is currently doing well.

Keywords Lymphocytic hypophysitis, Hypopituitarism

INTRODUCTION

Lymphocytic hypophysitis is the most common form of hypophysitis; the cause is usually unknown. It is initially characterized by lymphocytic infiltration and enlargement of the pituitary; this stage is followed by destruction of the pituitary cells.[1-3]

The clinical presentation of this inflammatory condition often mimics that of a pituitary adenoma.[4] It includes headache, vomiting, fatigue, hypopituitarism and diabetes insipidus. If there is mass effect on the optic chiasm, vision may be impaired, usually with bitemporal hemianopia, and if the cavernous sinus is involved, patients may have diplopia and orbital pain.[5]

This condition was initially thought to be an autoimmune phenomenon occurring primarily in women in the postpartum state. It is now recognized as a disorder that can affect both men and women over a rather large age range, from young adults to the elderly, and may be linked to a number of autoimmune phenomena.[6]

We report a case of a 45 years old male patient with acute adrenal crisis complicating chronic hypopituitarism secondary to lymphocytic hypophysitis.

CASE REPORT

A 45 years old male, with no previous comorbidities, presented to medical emergency in altered sensorium, with a history of severe holocranial headache associated with profuse vomiting of 4 days duration. Patient had a history of on and off low-grade fever for the past two months, associated with easy fatigability, anorexia, nausea, and weight loss with on and off periods of excessive vomiting. Patient's wife said that her husband had also noticed a reduction in his shaving frequency, and loss of axillary and pubic hair growth.

On examination, the patient was found to be disoriented (GCS-E4V2M6), afebrile, hypotensive, hypoglycemic, and in hyponatremia (Table 1). B/L pupils equal in size and reactive. No signs of meningeal irritation were present. No focal neurological deficits were observed.

Patient was managed with IV fluids, Dextrose and 3% NaCl for resuscitation of hypotension, hypoglycemia and euvolemic hyponatremia respectively, and was started on IV hydrocortisone on suspicion of adrenal crisis due to possible pituitary apoplexy. The patient improved clinically on conservative management. No history of polyuria, galactorrhea or erectile dysfunction/

impotence/decreased libido could be elicited. Visual acuity was normal in both eyes. No visual field defects or cranial nerve abnormalities seen.

Lab parameters are given in Table 1-further workup revealed the patient to have secondary adrenal insufficiency with secondary hypothyroidism and hypogonadotropic hypogonadism with low normal prolactin (adenohypophysis involved with predominant ACTH and TSH axis).

Table 1: Lab parameters.

Parameters	
WBC count-5300 cells	S. Na- 111 mEq/L, S. K- 4 mEq/L
Hb- 10 g/dl	Random blood sugar-30 mg/dl
Platelets-1.55 lacs	8 AM serum cortisol, <1 mcg/dl
Urea- 38 mg/dl	ACTH- <5 pg/ml
Creat-0.7 mg/dl	Free T3-1.98, free T4-1.08, TSH<0.003
ALT-91 IU/L, AST-146 IU/L, total bilirubin- 1.2 mg/dl	LH-1.33, FSH-2.30, total testosterone-38.86 ng/dl serum prolactin-4.34 ng/mL
ESR-60 mm/hr	HIV/HBsAg/anti-HCV-non reactive
CRP-0.9 mg/L	COVID-19 RT-PCR Negative

CSF analysis-cells- 5 cells, lymphocytic, sugar-90 mg/dl, protein-35 mg/dl, ADA-6 IU/L, CSF GeneXpert for MTB negative (was unremarkable for meningitis).

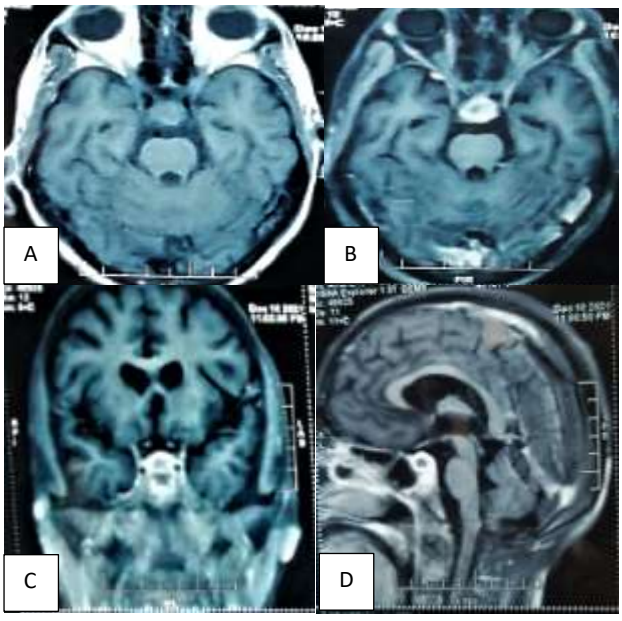

Figure 1 (A-D): Contrast MRI brain revealed homogeneous enlargement of anterior pituitary with upper portion convex, with thickened non- deviated lower infundibulum, showing peripheral enhancement of anterior pituitary with intact infundibulum, posterior pituitary and optic chiasma. No meningeal enhancement seen.

Contrast MRI brain (Figure 1) was done which revealed features suggestive of adenohypophysitis with intact pituitary stalk, cranial nerves, optic chiasma and posterior pituitary, and ruled out pituitary apoplexy.

Chest CT-no pulmonary infiltrates/hilar adenopathy-unremarkable for granulomatous disorders like tuberculosis and sarcoidosis

USG Abdomen-normal study.

Serum ACE and IgG4 levels were within normal range, and ANA and c-ANCA negative.

DISCUSSION

Hypophysitis can be categorized based on etiology as primary or secondary.[7] Primary hypophysitis can be further categorized based on histology as lymphocytic, granulomatous, xanthomatous, IgG4 related, or mixed and based on anatomy as lymphocytic adeno-hypophysitis, lymphocytic infundibuluneuro hypophysitis or lymphocytic panhypophysitis.[8,9]

Primary lymphocytic adeno-hypophysitis should be considered in the differential of any patient who presents with headaches of an intensity out of proportion to the size of the lesion and with hypopituitarism with preferential hypofunction of ACTH- and TSH-secreting cells, leading to adrenal insufficiency and hypothyroidism.[10] It affects women more than men with the average age of diagnosis for men being approximately 45 years and for women 34 years.[11]

The disease still remains a diagnosis of exclusion, and a definite diagnosis requires histopathological confirmation via transsphenoidal surgery, although a clinico-radiological diagnosis can be made in such settings where the conventional indications of surgery (mass effect, chiasmatic involvement, cavernous sinus invasion) are not present and surgery is not feasible.

MRI brain revealed homogenous enlargement of anterior pituitary with peripheral enhancement and non-deviated stalk which was isointense on T1 pre contrast images, which ruled out a pituitary adenoma with apoplexy. Further workup (serological and radiological) was unremarkable for infective causes (Tuberculosis), granulomatous disorders (sarcoidosis, granulomatosis with polyangiitis) and IgG4 related disease which constitute common secondary causes of hypophysitis.

Patient was managed on IV hydrocortisone (50 mg every 6th hourly) for a duration of 5 days, following which patient had significant improvement in blood pressure and sodium levels. Patient was discharged on hormone replacement therapy with 20 mg/day hydrocortisone and 50mcg levothyroxine (in the absence of significant mass effect and chiasma compression) and is currently doing better on follow-up.

CONCLUSION

This is an unusual case of acute adrenal crisis in a middle-aged male patient with chronic hypopituitarism due to primary lymphocytic adenohypophysitis. A high index of suspicion is of utmost significance since the inflammatory process often resolves after several months of glucocorticoid treatment, and pituitary function may be restored, depending on the extent of damage.

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