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Case Report Open Access

Post-partum Lymphocytic Hypophysitis in Ethiopian Woman

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Abstract

A 28 year-old young woman who was referred from Bahir Dar, Ethiopia for possible neurosurgical evaluation and management after she presented with headache and failure to lactate one month following delivery.The initial hormonal analysis was consistent with hypopituitarism.The magnetic resonance imaging (MRI) of the Brain done initially showed diffuse enlargement of the pituitary gland and thickening of the pituitary stalk. Most of the pituitary axes recovered after one month of hormonal replacement therapy.The follow-up MRI done after four weeks showed resolution of pituitary gland enlargement and stalk thickening.Based on clinical presentation, pituitary hormonal insufficiency and Brain MRI findings,the diagnosis of lymphocytic hypophysitis was made. Lymphocytic hypophysitis is a rare disease in which inflammation of the pituitary gland and stalk occurs. It is associated with transient or permanent insufficiency pituitary hormones. It usually occurs in women during third trimester of pregnancy or postpartum period. Systemic corticosteroid treatment at early stage may avoid unnecessary surgery in some cases but there are reported cases of spontaneous resolution.

Keywords: Hypopituitarism; Lymphocytic hypophysitis; Pregnancy; Postpartum period

Introduction

Lymphocytic hypophysitis is a rare disease characterized by inflammation of the pituitary gland and stalk. It is generally seen in women during third trimester of pregnancy or postpartum period [1].The estimated incidence of Lymphocytic hypophysitis is one case per 9 million people [2].The incidence reported might be underestimated because of the recent recognition of IgG4 related disease and immune check point inhibitor related disease [3]. It is often associated with other autoimmune disease conditions like Autoimmune thyroiditis, Addison's disease and Systemic lupus erythematosus.The histological characteristics show lymphocytic infiltration accompanied by edema and fibrosis with different levels of severity that may lead to transient or permanent hormonal dysfunction [4].

Patients with hypophysitis present with symptoms related to mass effect from pituitary gland enlargement and dysfunction. The area the pituitary gland affected and the size of its enlargement determine the various clinical manifestations. The treatment of Lymphocytic hypophysitis has not been examined well in prospective controlled studies. Medical management using immunosuppressive drugs can induce remission of autoimmune hypophysitis by treating both pituitary dysfunction and compression symptoms [5]. For those with mass effect like Visual loss, intractable headache and lack of response to medical therapy may require decompressive transsphenoidal surgery. In a number of published case reports of lymphocytic hypophysitis pituitary enlargement that resolved spontaneously has been described. In one group of study done recently showed radiologic finding regression in 15/15 patients receiving supportive therapy. Here we describe a patient in the postpartum period with markedly enlarged pituitary gland due to lymphocytic hypophysitis and associated hypopituitarism on replacement dose of corticosteroid with follow up clinical and radiologic improvement.

Case Report

A 28 year-old young woman who was referred from Bahir Dar, Ethiopia for possible neurosurgical evaluation and management after she presented with headache and failure to lactate one month following delivery. The headache was severe global in type with difficulty opening her eyes. It was unresponsive to analgesics and stayed for six consecutive days with subsequent resolution. She gave birth at term through caesarian section the indication being failed induction and the outcome was an alive male neonate weighing 3.6kg. There was no history suggestive of obstetric hemorrhage. She has no visual impairment and started seeing her menustration regularly following delivery.

Initial Brain MRI showed diffuse enlargement of the pituitary gland measuring 16x15x14mm and thickening of the pituitary stalk measuring 5.7 mm which suggests lymphocytic hypophysitis [Figure 1]. Patient was initiated on hormone replacement therapy with Prednisolone 7.5 mg PO daily for secondary adrenal insufficiency, and followed by Levothyroxine 25 mcg PO per day for central hypothyroidism. After four weeks of follow up treatment, she showed marked clinical improvement with no headache and started to breast feed her infant. She has also improvement in hormonal axes . Control Brain MRI showed resolution of pituitary gland enlargement measuring 9x8x5mm and pituitary stalk thickening measuring 2.9 mm. Progressively her prednisolone was reduced to 5mg PO daily and levothyroxine discontinued after normalization of the free thyroxine level.

Table 1. Physical examination	
Gender	Female
Age	28
Blood Pressure [in mmHg]	110/72-120/80
Pulse rate[beats/min]	74-80
Thyroid examination	Normal
Lymph nodes	Normal
Chest examination	Normal
Cardiovascular examination	Normal
Neurologic examination	
Consciousness	Alert
Visual acuity	6/6
Visual field	Normal
Fundus ophthalmoscopy	Normal
Cranial nerve examination	Normal
Sensory examination	Normal
Motor examination	Normal

Table.1: Table showing Physical examination.

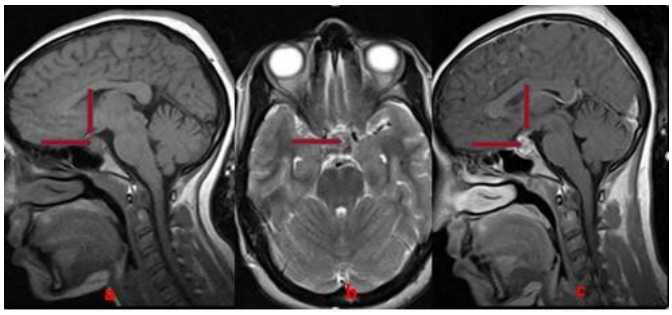

Figure 1. Initial MRI (aT1weighted sagittal Brain MRI (b T2 weighted axial Brain MRI (c T1weighted post contrast sagittal Brain MRI; diffuse enlargement of the pituitary gland (right arrow measuring 16x15x14mm and thickening of the pituitary stalk (down arrow which show T1 isointense signal and T2 mildly hypointense signal to the cortex; and showing heterogenous avid diffuse enhancement on contrast material enhanced T1 sagittal Brain MRI. These features suggest lymphocytic hypophysitis.

Discussion

Lymphocytic hypophysitis is more common in female than male in a ratio of 6:1 according to P. caturegli, et al, and tend to occur at younger age in women [35 ± 13 years] compared to men [45 ± 14years] that is consistent with our case. It is found that significant percentage of cases of lymphocytic hypophysitis occur during third trimester of pregnancy and the first two months in the post-partumperiod, which is similar with our patient who presented one month after delivery. One of the common presenting symptoms in the "single center case series" described by Brandon S. Imber et.al, was headache that was found in about 57% of cases and it was the main presentation in our case. In about 80% of cases multiple hormone deficiencies are found in lymphocytic hypophysitis which is a presentation in our case with cortisol, thyrotropin and gonadotropin deficiencies. Inhibition of lactation can be found in about 11% of case.Failed breast feeding was one of the first presentation in our patient possibly associated with relative hypoprolactinemia. Due to pituitary stalk compressive effect some cases may present with mild hyperprolactinemia which is not found in our case. Our patient's initial presentation of severe headache with difficulty opening the eyes and unresponsiveness to analgesics made us to consider pituitary apoplexy as a possible differential diagnosis as it could present with similar signs and symptoms in the post- partum period. In our case the improved clinical and hormonal responses and the marked Brain MRI normalization to replacement dose of steroid avoided unnecessary neurosurgical management. According to the description made by S.khare et al ,characteristically cortcotrophs are affected in about 75% of cases lymphocytic hypophysitis but in pituitary adenoma the last hormone to be affected is corticotroph which was the case in our patient with secondary adrenal insufficiency resulted from hypophysitis.

Brain MRI is the imaging procedure of choice in the diagnosis of lymphocytic hypophysitis. currently it is not easy to distinguish specifically hypophysitis from a pituitary adenoma based on radiologic finding and about 40% of patients are misdiagnosed as having pituitary macroadenoma and undergo unnecessary surgery. Therefore,based on the presence of clinical features and radiologic signs a scoring system by Gutenberg, A.,et al was proposed. .Here we

applied the scoring to our patient individual items were added and a score of −12 was obtained which is suggestive of lymphocytic hypophysitis .

Lupi et al. reviewed a total of 44 cases of autoimmune hypophysitis treated with glucocorticoids and/or azathioprine. A reduction in the size of the adenohypophysis was found in 84% of patients, the function of the adenohypophysis improved in 45% and that of neurohypophysis in 41% of these patients.In our patient the majority of the hormone deficiency and all the symptoms had improved.The sizes of the pituitary and stalk were normalized following one month treatment with corticosteroid indicating the underlying autoimmune etiology of lymphocytic hypophysitis and also showing that other inmunosupressive drugs can be used in the management of these cases. The role of corticosteroids in improving compression symptoms and as a replacement in lymphocytic hypophysitis is helpful in avoiding unnecessary transsphenoidal surgery. Even though pituitary tissue biopsy was not done, on the basis of marked clinical response and MRI normalization following to glucocorticoid replacement strongly suggests lymphocytic hypophysitis. There are different explanations why lympocytic hypophysitis occurs during the peripartal period, according to O'Dwyer, et al described the strong association of pregnancy and pituitary autoimmunity with the production of pituitary autoantibodies by patients that recognize a target autoantigen protein identified as alpha-enolase from the placenta. They also described another antibody, the neuronal specific enolase (NSE) which is expressed in both pituitary and placenta cell that establishes the direct link between pituitary and placental autoantigens. This gives a theoretical basis for the occurrence of lymphocytic hypophysitis in the peripartum period though it needs further verification with more studies.

Conclusion

Lymphocytic hypophysitis should be considered in the differential diagnosis of women who present with pituitary enlargement during late pregnancy and post- partum period.Close clinical evaluation and monitoring for multiple hormone deficiencies is required. Early treatment with pulse steroids may avoid unnecessary surgery. It is suggested that long-term follow-up with hormonal and MRI studies are necessary.

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