Postpartum Hypophysitis: A Diagnostic Pitfall

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ABSTRACT

We report an exceptional case of lymphocytic hypophysitis in a woman who was nine months pregnant. Despite the use of MRI, preoperative diagnosis remained difficult. Surgery was required after delivery due to worsening visual disturbances. The postoperative course was favorable, with a marked improvement in visual acuity and normalization of the follow-up brain MRI. This condition remains poorly understood despite evidence supporting an autoimmune disease.

Keywords: Hypophysitis, Pituitary gland, Endonasal surgery.

INTRODUCTION

Lymphocytic hypophysitis is a rare and recently described condition. The typically rapid onset of endocrine disorders, often associated with visual disturbances, are frequently the first clinical signs. While not pathognomonic, their occurrence, most often in pregnant women, poses difficult diagnostic and therapeutic challenges, justifying a renewed discussion of this unusual disease.

Case Report

A 34-year-old pregnant woman, with no significant past medical history, G4P3, presented at 37 weeks of amenorrhea with a sudden decrease in visual acuity in one eye.

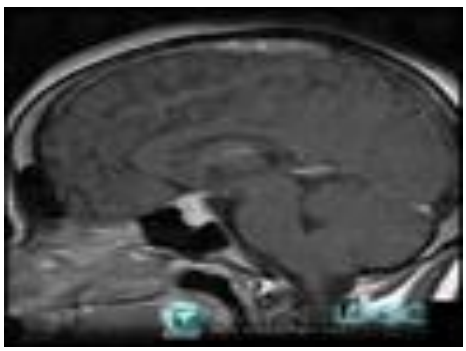

Fig.1:cerebral IRM en T2 (sagittal section)

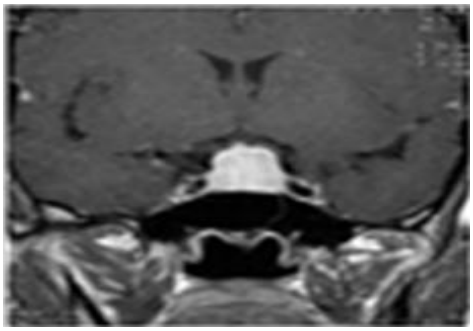

Fig.2: cérébral IRM en T2 (coronal section)

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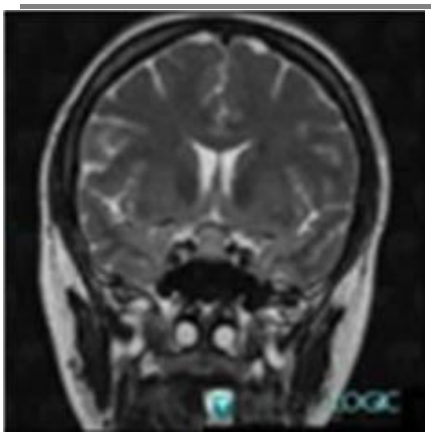

Fig.3: cérébral IRM en T1 (coronal section)

Visual acuity was measured at 5/10 in the left eye and 10/10 in the right, with a normal fundus examination [Fig 4].

Hormone	Normal	patient
FSH	3-9mUL/ml	0.9mUL/ml
LH	2-10mUL/ml	0.3mUL/ml
CORTISOL	250-700nmol/l	950nmol/l
PROLACTIN	0-20ng/ml	150ng/ml

Fig. 4 Hormonal panel

An orbital and brain MRI was performed, revealing a cystic, fluid-filled lesion, hypointense on T1-weighted images and hyperintense on T2-weighted images, measuring approximately 30 x 20 mm along its longest axis. This lesion occupied the sella maxillary fossa and extended superiorly to the optic chiasm cistern, causing displacement and superior displacement of the optic chiasm [Figs. 1, 2, 3]. Gadolinium injection was not administered due to the ongoing pregnancy.

Two weeks postpartum, the patient returned with worsening visual disturbances.

The ophthalmological examination revealed visual acuity of 4/10 in the left eye and 2/10 in the right, bitemporal hemianopsia on visual field testing, and bilateral optic disc edema on fundus examination. Endocrine and laboratory tests were performed and came back normal. The patient was hospitalized for decompressive surgery.

The patient underwent surgery via a subfrontal approach [this surgical approach was selected to provied complete and immediate decompression of the optic nerves] The cystic fluid was clear, like spring water, and was drained. A few membrane fragments were taken for histological analysis. In the immediate postoperative period, the patient reported complete resolution of her symptoms. The histopathological examination showed the presence of an inflammatory infiltrate composed of lymphocytes, plasma cells, and a few eosinophilic polymorphonuclear leukocytes. The diagnosis of lymphocytic hypophysitis was made.

The patient was seen again for a follow-up appointment one month later. Her ophthalmological examination revealed visual acuity of 10/10 in both eyes, and her follow-up brain MRI was unremarkable [Figs. 5, 6, 7]. Her endocrine workup revealed mild hypocorticism. The patient was then referred to endocrinology for possible correction of her hypocorticism and for investigation of other autoimmune diseases.

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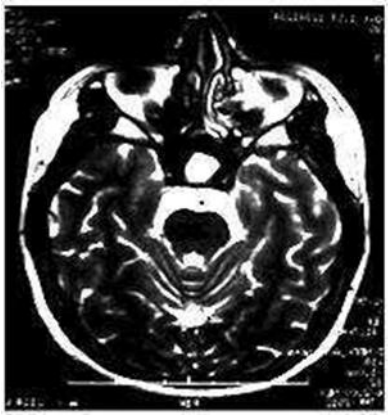

Fig.5: cerebral IRM en T2 (axial section)

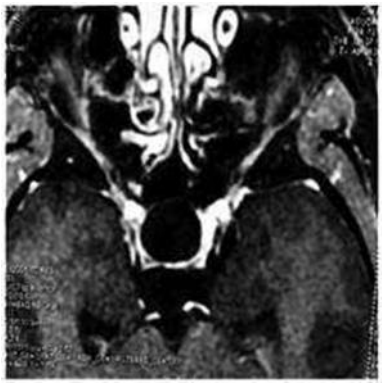

Fig.6: cérébral IRM en T1 (axial section) .

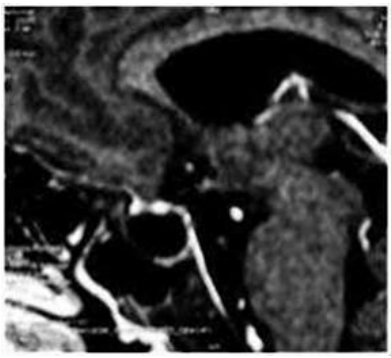

Fig.7: cérébral IRM en T1(sagittal section)

DISCUSSION

Lymphocytic hypophysitis is a rare and recently described entity. Since the first autopsy case described in 1962 by Goudie and Pinkertoh [10], approximately one hundred cases have been reported in the literature.

Lymphocytic hypophysitis is described in 72 cases in pregnant women or in the immediate postpartum period [1,3,4,5,11,12,15,14,18,20]. In 13 cases, it was discovered outside the context of pregnancy: multiparous women, women not pregnant [5,15], or menopausal women [7,8]. Two cases involving craniopharyngioma have been reported [17].

Three clinical signs may be indicative: the onset of headaches, associated with visual disturbances, as in Our patient [1, 4, 10, 18, 21] and/or endocrine disorders [17].

Laboratory tests are difficult to interpret during pregnancy; the results are generally normal, as in our case, although sometimes TSH and ACTH levels are slightly low [11, 15].

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A brain CT scan reveals an intrasellar mass with a more or less significant suprasellar extension responsible for the visual disturbances. This mass enhances with contrast, most often homogeneously, suggesting an enlarged pituitary gland. The question then becomes whether this is a common occurrence during pregnancy, an adenoma, or another pituitary pathology. A definitive diagnosis at this stage remains difficult; it can be suspected based on a cluster of findings [20b]: pregnancy, visual disturbances, and an intrasellar mass. Observations from brain MRI, as in our patient, show a regular increase in pituitary size with iso- or hypointense signal on T1-weighted images; it is not possible to identify an intrapituitary mass [18,20]. The mass appears hyperintense on T2-weighted images, and gadolinium injection results in homogeneous enhancement.

Several etiological hypotheses can be considered: inflammatory granuloma, tuberculosis, sarcoidosis, or adenoma, but the diagnosis of lymphocytic hypophysitis can only be made based on clinical examination or additional radiological investigations.

In all cases, a surgical procedure, at a minimum a biopsy, is necessary to perform a histological examination to obtain a definitive diagnosis. The timing of this biopsy depends on the clinical symptoms. If the disorders If visual symptoms worsen, as in our case, diagnostic and therapeutic surgery will be required quickly. In other situations, it is primarily a diagnostic procedure; this excisional biopsy is usually performed via a low endonasal or rhinoseptal approach, except when the extension is predominantly suprasellar [21], as in our case.

Histological examination in cases of lymphocytic hypophysitis shows stereotypical lesions: in all the cases described, as in our case, there is significant lymphocytic and plasmacytic infiltration of the pituitary gland [1,3,4,7,8,10,13,15,20,21]. Eosinophilic polymorphonuclear leukocytes are sometimes associated [3, 10], as in our case, as well as histiocytes.

The histological appearance easily rules out the diagnosis of adenoma. It should be noted, however, that an association between adenoma and lymphocytic pituitary is possible [14], even with a craniopharyngioma [17]. In the absence of histiocytic infiltration, idiopathic giant cell granulomatous hypophysitis, sarcoidosis, tuberculosis, or other (specific) conditions, as well as histiocytosis X [2,7], can be ruled out.

The presence of lymphocytes has led to lymphocytic hypophysitis being considered an autoimmune disease [19]. The association with other autoimmune diseases strengthens this hypothesis, such as Hashimoto's thyroiditis [10], anemia, sarcoidosis [11], and adrenal insufficiency [10].

Immunohistochemically, the lymphocytic infiltrate is polymorphic, often with a predominance of T lymphocytes [10]. The plasma cell population is polyclonal and A predominance of IgG plasma cells is often observed [10].

Before surgery, the diagnosis is rarely made, but it may be suspected [20] because not all pituitary masses during pregnancy or postpartum are adenomas [16].

Surgical access to these lesions involves a low endonasal or rhinoseptal approach. Subtotal resection with decompressive intent is recommended, leaving healthy tissue to avoid permanent hypopituitarism.

Before surgery, medical treatment with dexamethasone may be offered [20].

In our patient, this treatment was not prescribed due to the threat to visual function.

The search for anti-pituitary antibodies should be systematic, even if the results are often negative. Indeed, these patients must be followed regularly to determine whether they will develop an autoimmune disease in the future.

CONCLUSION

The possibility of diagnosing lymphocytic hypophysitis in pregnant women should be considered. Surgical intervention has a dual purpose: definitive diagnosis and therapeutic decompressive treatment, while avoiding complete resection.

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The increasing number of observed cases may lead to a revision of the anatomopathological classification of these pituitary inflammatory phenomena.

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