

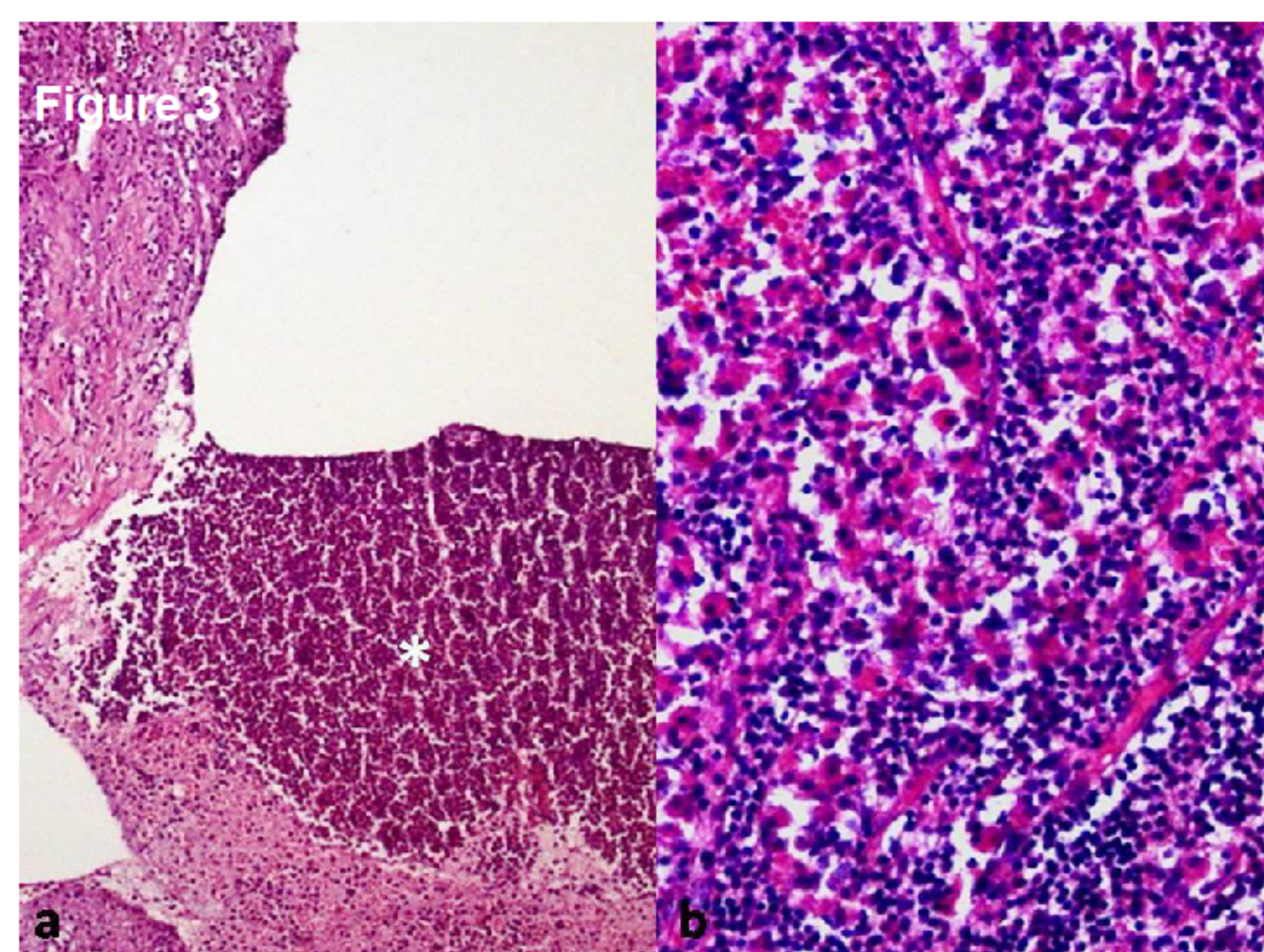
A rare adventure of a lymphocytic hypophysitis

Zeynep Cantürk, Alev Selek, Berrin Çetinarslan, İlhan Tarkun, Zeynep Akyay, Burak Çabuk, Savaş Ceylan

Kocaeli University Faculty of Medicine, Department of Endocrinology and Metabolism
Kocaeli University Faculty of Medicine, Department of Neurosurgery

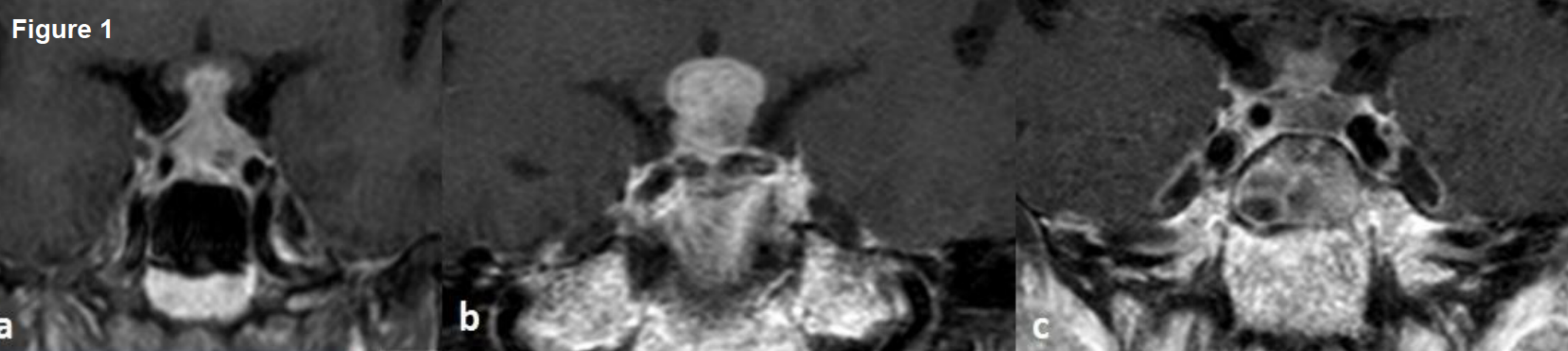
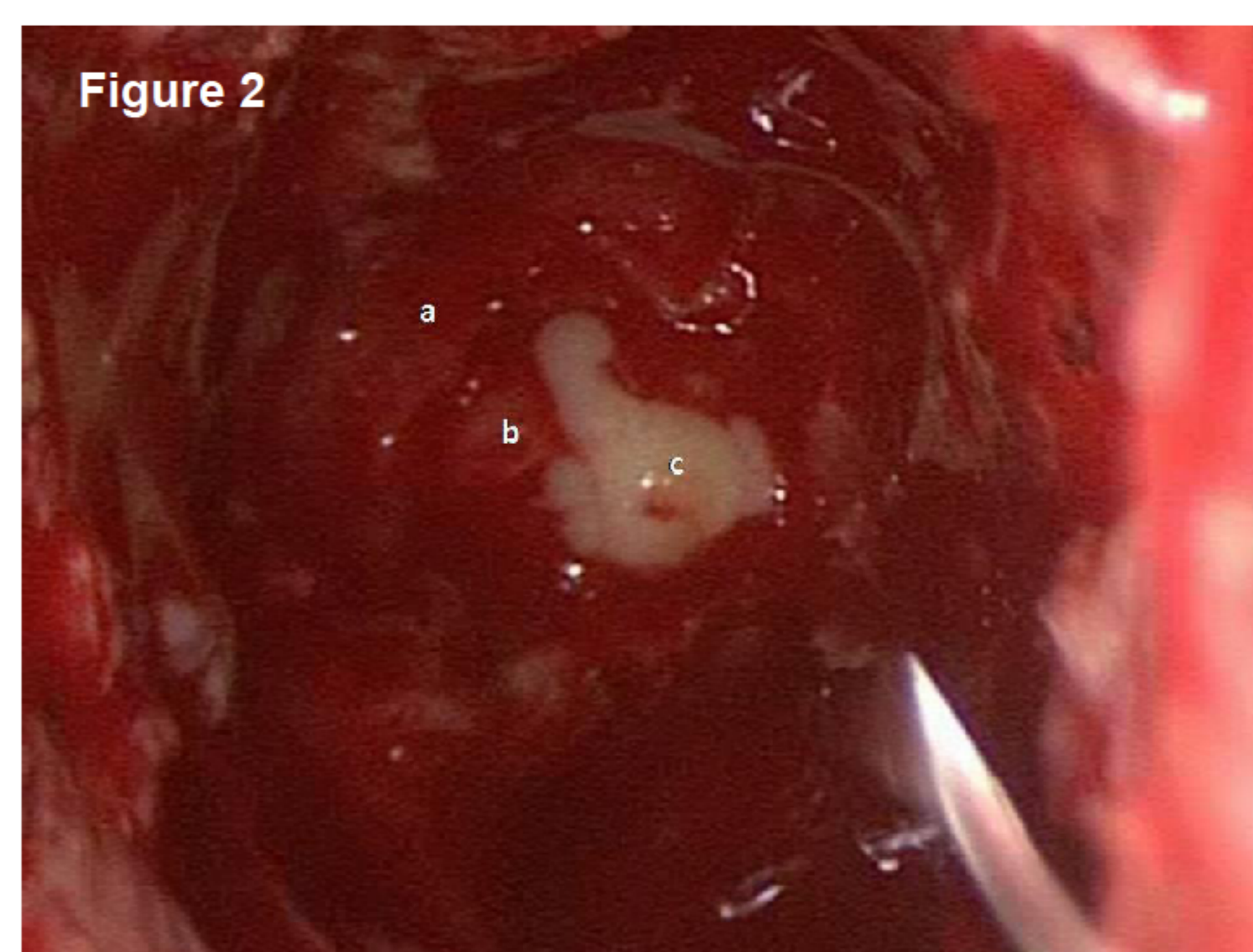
OBJECTIVES

Lymphocytic hypophysitis (LH) is a rare inflammatory disease of the pituitary gland. It is generally accepted as an autoimmune disease, because of the existence of antipituitary antibodies and concomitant autoimmune diseases, especially Hashimoto's thyroiditis.^[1,2] Definitive diagnosis relies on tissue biopsy.^[2] However, a homogenous enhancing sellar mass coincides with pregnancy, diabetes insipidus or hypopituitarism with or without hyperprolactinoma and a coexisting autoimmune disease may predict a presumptive diagnosis.^[3] Clinical course of the disease is a spectrum; from spontaneous resolution resulting empty sella, to recurrent invasive pituitary mass with severe compressive symptoms. This case is interesting for different aspects; as far as our knowledge, pituitary abscess formation secondary to preexisting lymphocytic hypophysitis has not been described before. On the other hand, *Staphylococcus Lugdunensis* has also not been described as a causative agent for pituitary abscess.



CASE

58 years old female was admitted to our hospital with one year history of headache and a pituitary adenoma. The pituitary lesion was 9mm in diameter on initial magnetic resonance imaging (MRI). Pituitary functions were normal (table 1) but during time course she had lost her anterior pituitary functions progressively resulting panhypopituitarism (table 1). On admission, pituitary MRI revealed a heterogeneous pituitary lesion, 14x11 mm in diameter with minimal peripheral contrast enhancement and thickened pituitary stalk (8mm) (figure 1a). According to clinical course and MRI findings, hypophysitis was the preliminary diagnosis. Trans-sphenoidal pituitary surgery was done and a purulent discharge was seen during surgery (figure 2). Gram staining showed gram positive coccus and *Staphylococcus Lugdunensis* was cultured. Antibiotherapy with meropenem and linezolid was initiated. Pituitary biopsy revealed pituitary abscess formation and lymphocytic infiltration of the remaining pituitary gland (figure 3a,b). Langerin, S-100 and CD1a immunohistochemical staining were all negative. IgG4/ IgG ratio was 1/10 and serum IgG4 levels were normal. LH with pituitary abscess was diagnosed after other causes were excluded. At the sixth month of follow up headache and fever were started again. On imaging, the pituitary lesion was relapsed but it was larger with extension to the hypothalamus and optic chiasm (figure 1b). There were bitemporal hemianopsia on visual field examination and second surgery was done in order to rule out recurrent pituitary abscess and decompression. There was no abscess formation and fibrotic tissue was biopsied. Intravenous infusion of 100 mg methylprednisolone for 3 days and oral administration of 60 mg/d prednisone for two weeks was started. At the end of second week of steroid treatment, toxic hepatitis was developed. Therefore the prednisone dosage decreased earlier and the entire treatment course lasted 12 weeks. At the first week of the treatment headache and fever was cured, her visual field was completely normal. 100mg/day azathioprine was started after normalization of her liver enzymes. The pituitary lesion reduced more than 50% on the third month MRI and she is still on follow up without any recurrence (figure 1c).



CONCLUSIONS

Lymphocytic hypophysitis may cause complete loss of vital pituitary functions and neurological deficits. The disease may rarely be complicated with abscess, therefore careful evaluation and surgical treatment should be performed in these cases. Radiologic features are variable from suprasellar extending mass to empty sella. Extrasellar disease is very rare but pituitary stalk thickening is seen in most cases. Our case presented with a sellar mass but the recurrent lesion was extending to suprasellar region, reaching to hypothalamus with optic chiasm compression. Definitive diagnosis requires histopathological evaluation. Microscopically, a diffuse infiltrate of inflammatory cells, mostly lymphocytes, is seen with nests of normal acinar cells surrounded by necrosis and extensive fibrosis. The first operative pathology of our patient revealed neutrophilic infiltration of the centre suggesting abscess formation and lymphocytic infiltration of the surrounding hypophysial cells therefore LH with pituitary abscess was diagnosed. Most symptomatic LH require pulse doses of methyl- prednisolone or prednisone followed by a slow taper over a period of weeks to months. Despite good response to these first-line therapies in the majority of patients, relapses are common. In recurrent and symptomatic lymphocytic hypophysitis, high-dose glucocorticoids combined with azathioprine may be a successful choice of treatment. As a conclusion; LH may rarely be complicated with abscess, this is not a simple coincidence. The disrupted pituitary tissue might become susceptible to infections.

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