A Case of Recurrence of Congenital Ocular Toxoplasmosis with Frosted Branch Angiitis (Ocular Toxoplasmosis with Frosted Branch Angiitis)

Takahiro SUZUKI, Hiromi ONOUCHI Yoshihiro NAKAGAWA, Hideki OOHASHI, Han KAIKEN and Kenji KAWAI

Department of Ophthalmology, Tokai University School of Medicine

(Received January 6, 2010; Accepted August 2, 2010)

Purpose: To describe a case of recurrence of congenital ocular toxoplasmosis with frosted branch angiitis.
Case Report: A 24-year-old woman presented with hyperemia in her right eye. Medical history included epilepsy at age 14 and mild mental retardation. Iridocyclitis and vitreous opacity were observed in the right eye, and furthermore widespread retinal vessel sheathing due to frosted branch angiitis was seen. Acyclovir was initiated for acute retinal necrosis with frosted branch angiitis. One week later, serologic tests showed elevated toxoplasma antibody level and toxoplasma antibody IgG level, and a white retinal exudative lesion with unclear margins was noted. Therefore, acetylspiramycin and prednisolone were initiated for a recurrence of congenital ocular toxoplasmosis. After treatment, inflammation subsided, the exudative lesion shrank, and the frosted branch angiitis improved.
Conclusion: We encountered a case of ocular toxoplasmosis due to recurrence of congenital toxoplasmosis with frosted branch angiitis. The clinical symptoms of ocular toxoplasmosis can be varied and the diagnosis should be kept in mind.

Key words: recurrence of congenital toxoplasmosis, frosted branch angiitis

INTRODUCTION

Ocular toxoplasmosis, which is caused by intracellular parasitism by Toxoplasma gondii, is one of the main causes of uveitis [1]. Frosted branch angiitis was originally reported by Ito in 1976 in a six-year-old child presenting with severe sheathing of all retinal vessels producing the appearance of frosted branches of a tree [2]. Although periarteritis related to toxoplasmosis is common, frosted branch angiitis has been infrequently described in patients with toxoplasmic retinochoroiditis [3]. In this study, we report a case of recurrence of congenital ocular toxoplasmosis with frosted branch angiitis.

CASE REPORT

A 24-year-old woman visited a regional hospital with hyperemia in her right eye. She consulted our hospital the following day for further examination and treatment. Her social history was unremarkable. Her past Medical history included epilepsy at age 14 and mild mental retardation. At our initial ocular examination, visual acuity was 0.03 in right eye and 1.2 in left eye. Intraocular pressure was 17 mmHg in right eye and 14 mmHg in left eye. Iridocyclitis, vitreous opacity, and extensive white sheathing of retinal vessels were seen in the right eye, and there were 2+ cells in the anterior chambers and vitreous humor in one's eye. Examination of the retina in the posterior pole showed intense vitreous opacity and sheathed blood vessels in the right eye (Fig. 1a). The retina in the periphery, the vessel sheathing and retinal exudates also was shown (Fig. 1b).

Fluorescein angiography showed dye leakage from the vessels with sheathing, but other details were unclear due to vitreous opacity (Fig. 2a, b). Ophthalmic evaluation of the left eye revealed normal results. Routine laboratory evaluation, including complete blood count, blood chemistry, urinalysis, chest x-ray, and serologies for toxoplasma, human immunodeficiency virus, and syphilis, were performed. First, according to clinical findings, acyclovir was initiated for probable acute retinal necrosis with frosted branch angiitis. One week later, the results of peripheral blood tests and blood biochemistry tests showed a mild rise in leukocyte count, and Antitoxoplasma antibodies (latex agglutination assay: LA) and toxoplasma IgG antibodies (indirect fluorescent antibody technique: FA) were positive (1,280- and 114-fold, respectively). Other serologic test results, including antitoxoplasma IgM antibodies (FA), Angiotensin-converting enzyme, and viral antibodies such as varicella zoster virus and herpes simplex virus were within normal limits. In the retina, scar-like lesion and a white retinal exudative lesion with unclear margins were noted, and fluorescent fundus angiography findings included a hypofluorescent area at the scar-like lesion and a coinciding hyperfluorescent area at the exudative lesion (Fig. 3a, b). Therefore, as ocular toxoplasmosis, administration of acetylspiramycin 1,200 mg/day, pulse therapy with corticosteroids (Solu-Medrol 1,000 mg, intravenous infusion for 3 days), and subsequent oral prednisolone 30 mg were initiated. After treatment, iridocyclitis and vitreous opacity gradually resolved, and remission of frosted branch angiitis was seen. Six months later, visual acuity improved to 1.0, and subjective symptoms

Takahiro SUZUKI, Department of Ophthalmology, Tokai University School of Medicine, 143 Shimokasuya, Isehara, Kanagawa 259-1143, Japan
Tel: +81-463-93-1121 Fax: +81-463-91-9328 E-mail: youta@is.icc.u-tokai.ac.jp

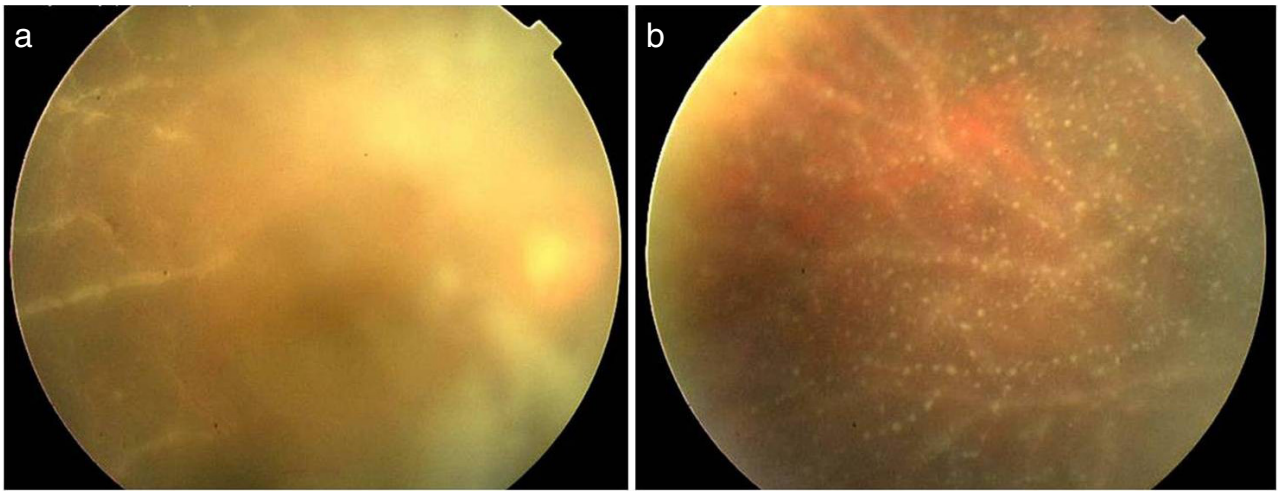

Fig. 1a, b Right fundus photography performed during the first visit showed intense vitreous opacity of the posterior pole and sheathed blood vessels. The vessels in the surrounding area also showed sheathing and retinal exudative macular findings.

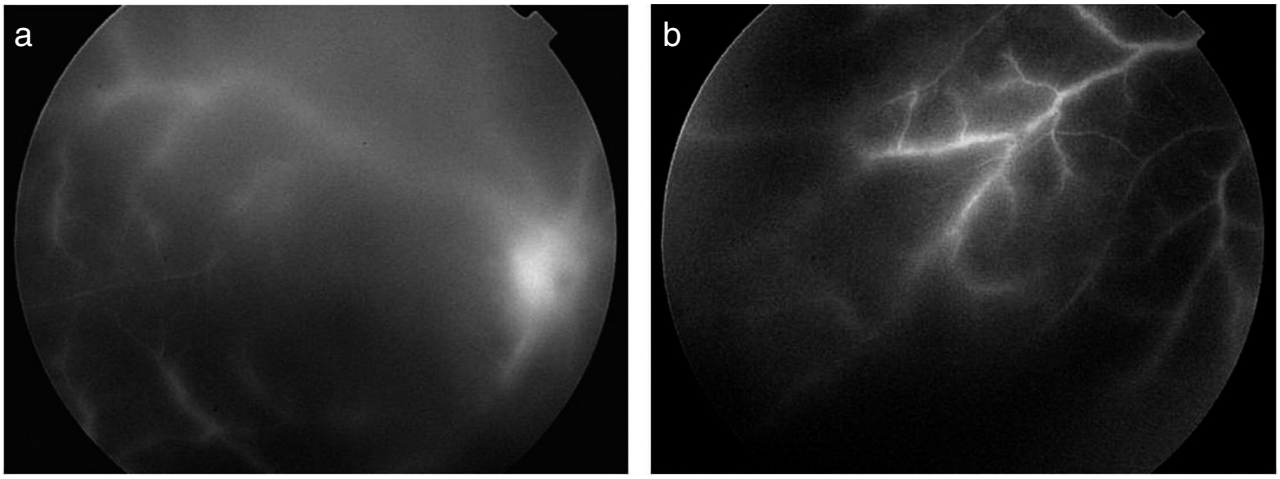

Fig. 2a, b Fluorescent fundus angiography of the right eye during the first visit showed fluorescent leakage from retinal vessels in both the posterior pole and the surrounding area on fluorescent imaging.

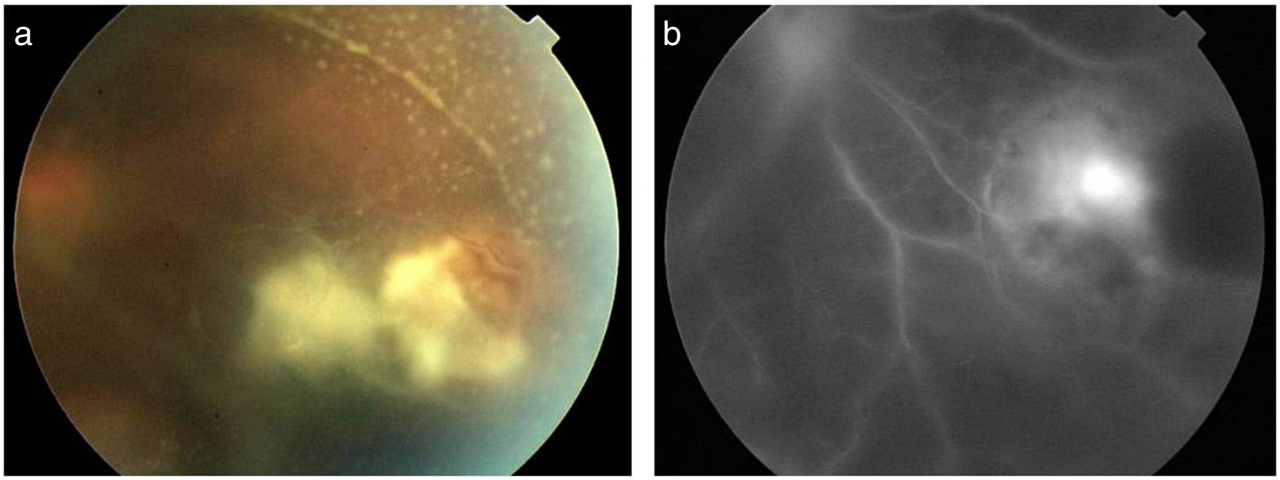

Fig. 3a, b Right fundus photography and fluorescent fundus angiography one week after the first visit showed a scar-like lesion on the nasal side below the papilla and a white retinal exudative lesion adjacent to it on fundus photography. On fluorescent fundus angiography, the scar-like lesion is hypofluorescent and the retinal white lesion is hyperfluorescent.

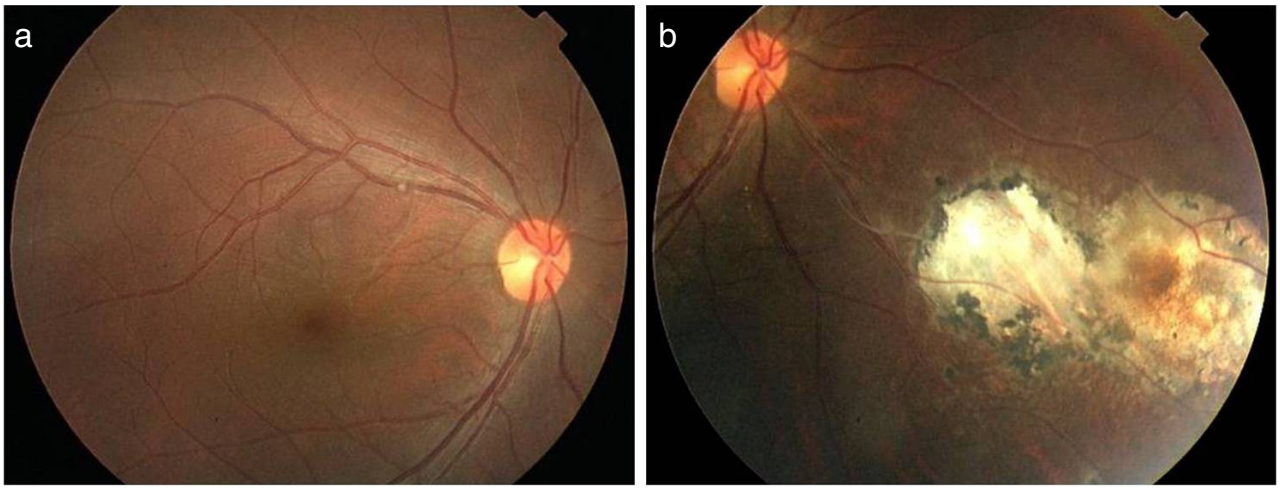

Fig. 4a, b Right fundus photography performed 6 months after the first visit showed remission of frosted branch vessels and necrotic scarring of the lesion.

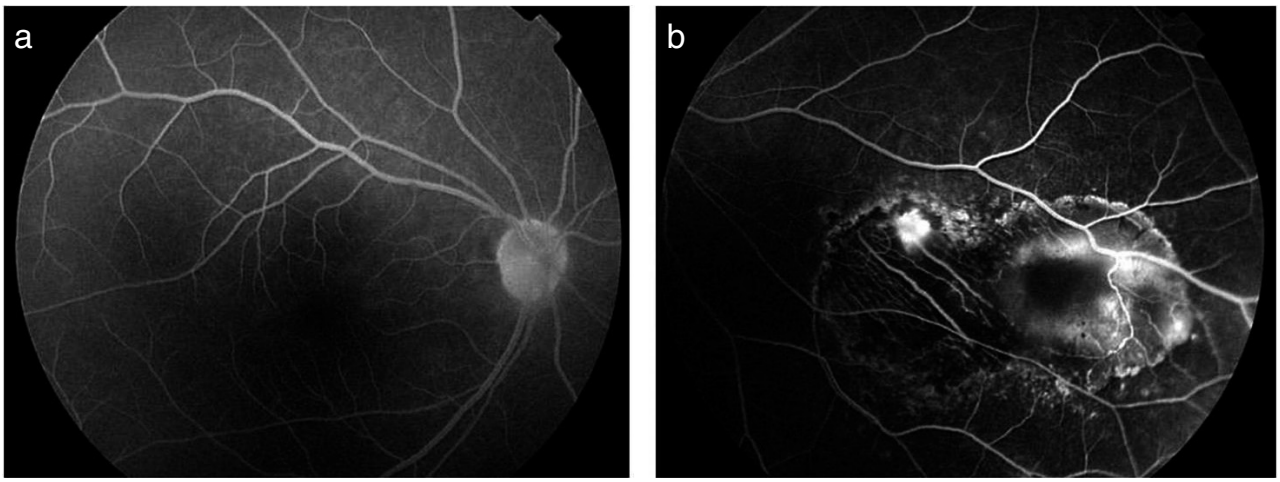

Fig. 5a, b Fluorescent fundus angiography performed 6 months after the first visit showed hyperfluorescence due to autofluorescence and hypofluorescence due to pigment proliferation, which was believed to be a residual lesion.

improved. The scar-like lesion and exudative lesion observed during the course of the disease manifested a necrotic cicatricial lesion with clear margins (Fig. 4a, b). Fluorescent fundus angiography showed hyperfluorescence due to autofluorescence and hypofluorescence due to pigment proliferation (Fig. 5a, b), and it transformed into a residual lesion.

DISCUSSION

Toxoplasma gondii infects a large proportion of the world's human populations, it is an uncommon cause of disease. Certain individuals, however, are at high risk of severe or life-threatening disease due to this parasite. These include congenitally infected fetuses and newborns and immunologically impaired individuals. Congenital toxoplasmosis is the result of maternal infection acquired during gestation, an infection that is most often clinically inapparent. In immunodeficient patients, toxoplasmosis most often occurs in persons with defects in T cell–mediated immunity such as those receiving corticosteroids, anti–tumor necrosis factor (TNF) therapies, or cytotoxic drugs and those with hematologic malignancies, organ transplants, or acquired immunodeficiency syndrome (AIDS). In the vast majority of otherwise immunocompetent individuals, primary or chronic (latent) infection with Toxoplasma gondii is asymptomatic; after the acute infection, a small percentage has chorioretinitis, lymphadenitis, or, even more rarely, myocarditis and polymyositis. *T. gondii* was first observed in the North African rodent (*Ctenodactylus gundi*) by Nicolle and Manceaux in 1908 and was recognized as a cause of human disease in an 11-month-old congenitally infected child by Janku in 1923. It was reported as a cause of encephalitis by Wolf and colleagues, who in 1939 observed it in a newborn who presented with seizures, intracranial calcifications, hydrocephalus, and chorioretinitis. Although relatively few cases of severe toxoplasmosis in adults were reported during the ensuing years, the remarkable report in 1968 by Vietzke and colleagues from the National Cancer Institute of the National Institutes of Health highlighted *T. gondii* as a cause of life-threatening infection in patients with malignancy, predominantly in those with hematologic malignancies. Brain involvement with focal areas of encephalitis was the primary finding at autopsy in these patients. Since that time, several hundred cases in non-AIDS immunodeficient patients have been recorded in the literature. In 1983, the first report of toxoplasmosis in AIDS patients appeared. Toxoplasmic encephalitis

(TE) subsequently was recognized as the major cause of space-occupying lesions in the brains of these patients, almost all of whom had serologic evidence of previous exposure to the parasite. Despite the significant advances that have been achieved in the recent past, major challenges remain in the areas of prevention and management of the acute infection in pregnancy, the fetus, and the newborn and in the understanding and treatment of toxoplasmic chorioretinitis and infection in immunocompromised individuals.

Ocular involvement is considered to be rare. [4−6] The present case was diagnosed as ocular toxoplasmosis secondary to recurrence of a congenital infection, based on the age of the patient, the medical history, the presence of a retinal exudative lesion, response to therapy specific for toxoplasmosis and serological findings.

The first reported case of frosted branch angiitis is said to be a case of bilateral sheathed retinal vessels in a normal young man in 1976. Since then there have been reports of typical retinal vessel findings and the efficacy of steroid preparations, but there have been few reports of ocular toxoplasmosis. This case is a rare case of frosted branch angiitis that occurred in conjunction with uveitis by ocular toxoplasmosis. We encountered a rare case of ocular toxoplasmosis considered to be recurrence of congenital toxoplasmosis with frosted branch angiitis. The clinical symptoms of ocular toxoplasmosis can be varied and the diagnosis should be kept in mind.

REFERENCES

1) McCannel CA, Holland GN, Helm CJ, *et al*. Causes of uveitis in the general practice of ophthalmology. Am J Ophthalmol 1996; 121: 35−46.
2) Ito Y, Nakano M, Kyu N, *et al*. Frosted branch angiitis in a child (in Japanese). Rinsho Ganka (Jpn J Clinic Ophthalmol) 1976; 30: 797−803.
3) Ysasaga JE & **Davis J. Frosted branch angitis with ocular toxo**-plasmosis. Arch Ophthalmol 1999; 1260−1261.
4) Akstein RB, Wilson LA, Teutsch SM. Acquired toxoplasmosis. Ophthalmology 1982; 1299−1302.
5) Montoya JG, Remington JS. Toxoplasmic chorioretinitis in the setting of acute acquired toxoplasmosis. Clin Infect Dis 1996; 23: 277−282.
6) Burnett JA, Shortt SG, Isaac-Renton J, *et al*. Multiple cases of acquired toxoplasmosis retinitis presenting in an outbreak. Ophthalmology 1998; 105: 1032−1037.