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Case Report
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Vogt-Koyanagi-Harada Disease Recurred after 8 Days of Initial Steroid Pulse Therapy: A Case Report

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Abstract

Purpose: This study aimed to report a case of Vogt-Koyanagi-Harada (VKH) disease that recurred 8 days after initial pulsed corticosteroid therapy, which is quite short.

Case and Findings: A 56-year-old male patient presented with blurred vision in both eyes and a headache for 6 days. His best-corrected visual acuity was 1.2 and 0.7 in the right and left eye, respectively. A serous retinal detachment in the posterior fundus was observed in both eyes. He was diagnosed with VKH disease. Fundus fluorescein angiography revealed multiple pinpoint leaks suggestive of VKH disease. He received 1000 mg of daily methylprednisolone infusion, for 3 days, followed by oral prednisolone, at 40 mg daily. Serous retinal detachment and headache promptly recurred during the oral administration of 40 mg of prednisolone, 8 days after initial steroid pulse therapy. Sub-Tenon triamcinolone acetonide was injected in both eyes, and serous retinal detachment and headache immediately improved. No recurrence was observed for 8 months until the present, and his best-corrected visual acuity was 0.9 in both eyes.

Conclusion: Sub-Tenon triamcinolone acetonide injection may be effective for VKH disease recurrence when added to conventional steroid pulse therapy. This case suggests the possibility of recurrence in the early stage of steroid pulse therapy.

Introduction

Vogt-Koyanagi-Harada (VKH) disease is an autoimmune-driven inflammation of the ocular, auditory, and meningeal structures that all contain melanocytes, which is the elective target of the inflammatory reaction [1,2]. VKH disease frequently recurs. Maruyama et al. reported that 20% of patients with VKH disease experience recurrence even with the best diagnostic and treatment strategies [3]. Several reports have been presented about the recurrence of VKH disease from 10 to 21 years after initial therapy according to a search of PubMed (keywords, 'VKH'; search years, 2000–2022) and CiNii Research (keywords, 'VKH'; search years, 2000–2022) databases [4-6].

On the contrary, Masaki et al. reported that VKH disease recurred 3 days after initial prednisolone high-dose therapy [7]. No reports were presented about the early recurrence of VKH disease following steroid pulse therapy through PubMed and CiNii Research. Hence, evidence is insufficient and details are unclear about the recurrence of VKH disease following steroid pulse therapy.

Herein, we present a case of serous retinal detachment type of VKH disease that recurred after 8 days of initial steroid pulse therapy. We report details in the current report as this case seems to be precious.

Case Presentation

A 56-year-old male patient presented at Dokkyo Medical University Saitama Medical Centre, Koshigaya, Japan in August 2022, with a headache and blurred vision for 6 days. He was suspected of VKH disease at a previous eye clinic. The Best-Corrected Visual Acuity (BCVA) was 1.2 and 0.7 in the right and left eye. Both eyes have normal intraocular pressure. Slit-lamp biomicroscopy revealed mild iritis, and cells in the vitreous cavity in both eyes. Fundus examination revealed serous retinal detachments in both eyes (Figure 1a). Fluorescein angiography revealed bilateral and multiple hyperfluorescence pinpoints at the posterior pole of the fundi (Figure 1b).

Optical coherence tomography demonstrated retinal pigment epithelium undulations and

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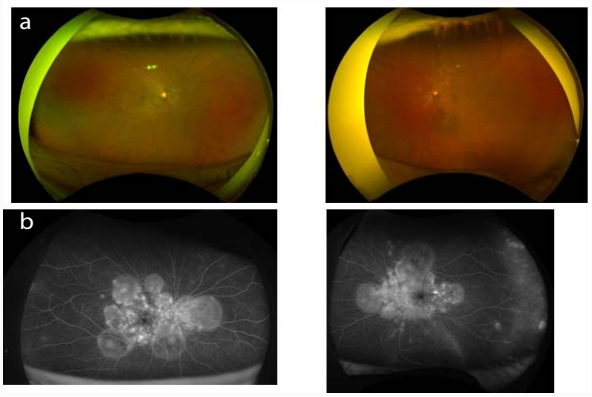

Figure 1: Eye examination images from a 56-year-old male patient with decreased vision in both eyes and a headache, showing (a) fundoscopy images showing serous retinal detachments in both eyes and (b) fluorescein angiography images showing hyperfluorescence, and multiple areas of pinpoint hyperfluorescent foci in the posterior pole of the fundi of both eyes.

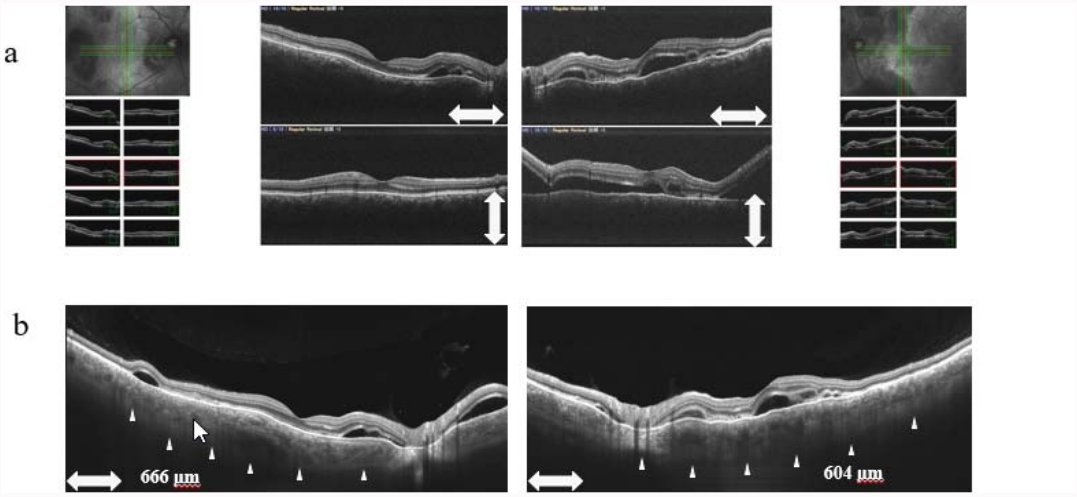

Figure 2: Optical coherence tomography images from a 56-year-old male patient with decreased vision in both eyes and a headache, showing (a) serous retinal detachments in both eyes and (b) choroidal thickenings in both eyes.

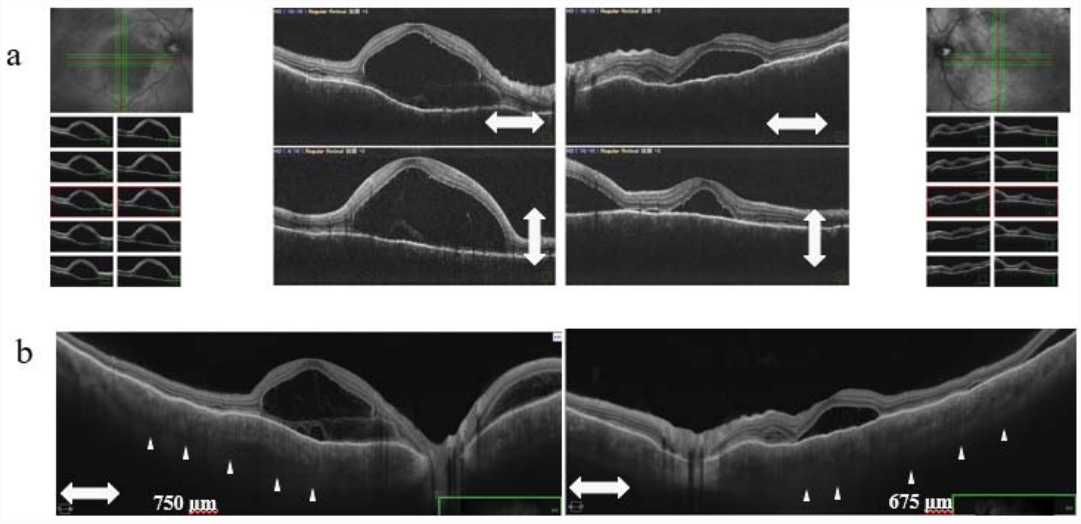

Figure 3: Optical coherence tomography images, 11 days after initial therapy from a 56-year-old male patient who presented with decreased vision in both eyes and a headache, showing (a) serous retinal detachments worsened in both eyes and (b) increased choroidal thickenings compared to the initial visit in both eyes.

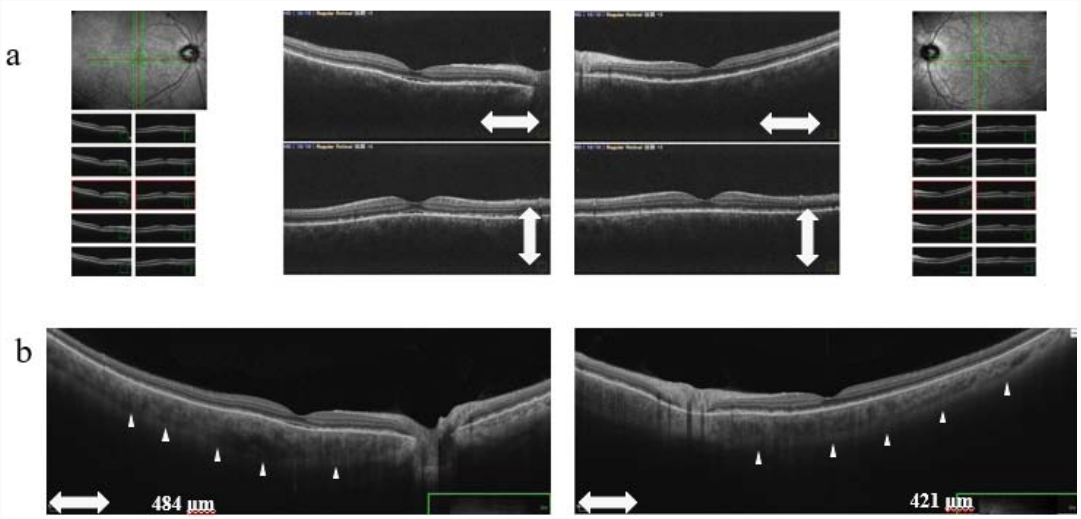

Figure 4: Optical coherence tomography images, 21 days after initial therapy from a 56-year-old male patient who presented with decreased vision in both eyes and a headache, showing (a) almost flat retinal pigment epithelium in both eyes and (b) reduced choroidal thickenings in both eyes.

serous retinal detachment (Figure 2a) and choroidal thickening (Figure 2b) in both eyes.

Blood test results were within normal limits, except for the red blood cell levels (4.23×10^{12} cells/l). The glycosylated hemoglobin (HbA1c) level was 5.3%.

The clinical diagnosis was VKH and the patient was immediately treated with 1000 mg of methylprednisolone (Solu-Medrol; Pfizer, New York, NY, USA) infusion daily for 3 days, followed by oral administration of 40 mg of prednisolone (Predonine; Shionogi, Osaka, Japan), daily, which was gradually tapered off over the subsequent 6 months.

Headache, retinal pigment epithelium undulations, and serous retinal detachment improved at first, and the patient was discharged from 7 days after initial steroid pulse therapy. Headache and decreased vision recurred 8 days after initial therapy. Serous retinal detachment worsened (Figure 3a) and choroidal thickness increased (Figure 3b) 11 days after initial therapy.

The BCVA was 0.2 and 0.5 in the right and left eyes, respectively. Sub-Tenon triamcinolone acetonide at 40 mg was injected in both eyes and serous retinal detachment and headache improved immediately. The BCVA improved by 0.6 in both eyes 21 days after initial therapy. Serous retinal detachment was almost resolved (Figure 4a) and choroidal thickness improved by 484 and 421 um in the right and left eyes, respectively (Figure 4b).

The therapeutic reaction was favorable, with no recurrence for 8 months until the present.

Discussion

One of the features of VKH is recurrence. Recurrence that repeats several times will become a chronic type. The visual prognosis of chronic type is generally unfavorable. The PubMed (keywords, 'VKH'; search years, 2000–2022) and CiNii Research (keywords, 'VKH'; search years, 2000–2022) databases, revealed that VKH disease recurred after 8 days of initial steroid pulse therapy, which is quite short. Ishibazawa et al. reported that headache, tinnitus, and sensorineural hearing loss developed, with increased choroidal thickness without other evidence of increased ocular inflammation after 6 months of initial steroid pulse therapy. Headache and ocular inflammation were observed in our case as recurrent symptoms. A few reports presented about the recurrence period from initial therapy, thus accumulation of cases is important.

Rubsamen et al. reported that VKH disease recurred in 9 (43%) of 21 patients in the first 3 months, usually in association with a rapid tapering of steroid dosage [9]. Iwahashi et al. reported that poor initial visual acuity and rapid tapering of the corticosteroid were associated with posterior recurrence [10]. Our patient recurred during an oral administration of 40 mg of prednisolone after 3 days of 1000 mg of methylprednisolone infusion. This change of medication may correspond to rapid tapering.

Hosoda et al. reported that sub-Tenon triamcinolone acetonide injection alone may have VKH recurrence [11]. Our patient received 40 mg of sub-Tenon triamcinolone acetonide injection added with oral administration of 40 mg prednisolone. Therefore, recurrence was not observed. Our treatment was effective not only in ocular findings but also in headache resolution.

T cell reactivation which recognizes melanocyte antigenicity, suppressor T cell hypofunction which restrains expression of antigenicity of melanocyte, and antigenicity decoration of melanocyte by virus infection seem to be the causes of the mechanism of VKH recurrence [4]. Immune reactions may occur for the target of residual melanocytes [4]. However, these assumptions are one of the speculations because the mechanism of VKH disease onset and recurrence remains unclear. Rapid elucidation is anticipated.

Conclusion

VKH disease may recur in an early stage of steroid pulse therapy from the current case. We have to pay attention to recurrence at any time. Sub-Tenon triamcinolone acetonide injection added to oral administration of prednisolone is quite useful for the recurrence of VKH disease.

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