Case Report

Bilateral Multiple Serous Retinal Detachments Following Bone Marrow Transplantation

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Abstract

Purpose: To describe a case of bilateral multiple serous retinal detachments (SRD) following bone marrow transplantation (BMT), which showed bilateral response to a single unilateral intravitreal bevacizumab injection.

Case Report: A 37-year-old man with acute myelogenous leukemia who had received bone marrow transplantation four months prior was referred to our clinic with the chief complaint of gradually decreasing vision in both eyes for three months. During the funduscopic examination, multiple serous retinal detachments (SRD) were observed bilaterally, and he was diagnosed with multiple foci of central serous chorioretinopathy (CSCR). He was advised to discontinue the steroid dosage, which did not make significant improvement, and he was treated with intravitreal bevacizumab injection in the more severely affected eye. One month later, significant improvement was noticed in both eyes.

Conclusion: Serous retinal detachment is a rare complication following BMT. Significant bilateral improvement after single unilateral intravitreal bevacizumab injection shows not only the possible role of increased level of vascular endothelial growth factor (VEGF) in this case, but also the systemic diffusion of the drug and effect on the contralateral eye following unilateral injection.

Keywords: Allogenic Bone Marrow Transplantation; Bevacizumab; Central Serous Chorioretinopathy; Serous Retinal Detachment; Vascular Endothelial Growth Factor

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INTRODUCTION

While ocular complications of allogenic bone marrow transplantation (BMT) are common, they most commonly present as anterior segment disorders including tear film abnormalities, keratoconjunctivitis sicca, corneal ulcer and cataract.[1] Posterior segment complications of

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BMT include microvascular angiopathy, fungal retinitis or endophthalmitis, cytomegalovirus retinitis,[2] and rarely central serous chorioretinopathy (CSCR).[3] Here we report a case of bilateral multiple serous retinal detachments (SRDs) following BMT in whom both eyes that responded significantly to a single unilateral intravitreal bevacizumab injection.

CASE REPORT

The patient was a 37 year-old man complaining of bilateral gradually decreasing visual acuity of three months' duration. His past ocular history was unremarkable, but he had a history of acute myelogenous leukemia (AML) for 1 year, and had received allogenic BMT 4 months prior to presentation to our clinic. His medications at presentation were oral prednisone 50 mg/day, cyclosporine 100 mg/day, and mycophenolate mofetil 1 g twice daily. During the ocular examination, his best corrected visual acuity (BCVA) was 20/50 in the right eye and 20/40 in the left eye. Afferent papillary defect was negative, and the anterior segment examination was normal in both eyes. In the fundus examination there were numerous foci of serous retinal detachment bilaterally. On fluorescein angiography (FA) multiple retinal pigment epithelium (RPE) leaking points were noted bilaterally [Figure 1] with gradual subretinal pooling in the retinal detachment areas [Figure 2]. In indocyanine green angiography (ICGA) images choroidal vessels were dilated in early phase [Figure 3] and choroidal hyperpermeability was present in late phase [Figure 4]. The extent of ICGA abnormalities were more severe than the clinical findings.

In ocular coherence tomography (OCT) images, there was intraretinal edema, and accumulation of subretinal hyporeflective fluid [Figure 5]. The RPE layer seemed to be intact.

He was diagnosed as having multiple bilateral CSCR and on consulting his internist, he was asked to discontinue the steroid completely. Two months later, no significant improvement was seen, so intravitreal bevacizumab was injected in his right eye, which had lower visual acuity and more subretinal fluid than the left eye. One month after the injection, BCVA was 20/30 in both eyes and in funduscopy there was marked improvement in the SRD areas bilaterally. FA [Figure 6] and OCT [Figure 7] also showed significant improvement in both eyes. The symptoms and clinical examination were stable 4 months after the injection.

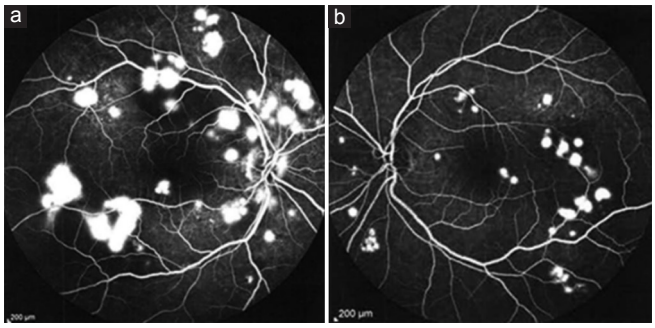

Figure 1. Fluorescein angiography of the right (1a) and the left eye (1b) at presentation: multiple leaking points with gradual subretinal pooling are visible in both eyes. Serous retinal detachment was present in the inferior part of the right eye (not visible here).

DISCUSSION

CSCR has been reported as a rare complication following allogenic BMT but its pathogenesis remains a topic of debate.[2-4] Corticosteroid usage is a well-known risk factor for development of CSCR after solid organ transplantation.[2-4] Lee et al in a retrospective study showed that reducing the steroid dose would accelerate remission of CSCR for about 2 months,[4] but in the current case, even discontinuing the steroid did not make significant improvement.

Other mechanisms unrelated to corticosteroids may also be responsible for the development of CSCR. Fawzi et al reported a case of multifocal bilateral CSCR following renal transplantation while the patient was receiving cyclosporine but no steroids, so they proposed that other mechanisms may be responsible in their case.[3]

Another plausible mechanism is choroidal microvascular compromise which has been reported in both graft versus host disease (GVHD),[5] and with cyclosporine use.[6] Cheng et al reported a case of bilateral CSCR following BMT which resolved with systemic high dose steroids.[5] They suggested that choroidal infiltration in GVHD has caused an increase in choroidal vessel permeability resulting in CSCR. In view of the fact that our patient was being treated with anti-GVHD prophylaxis and none of the more common ocular presentations of GVHD were present, this mechanism seems less probable., Cyclosporine has been shown to induce thrombotic microvascular

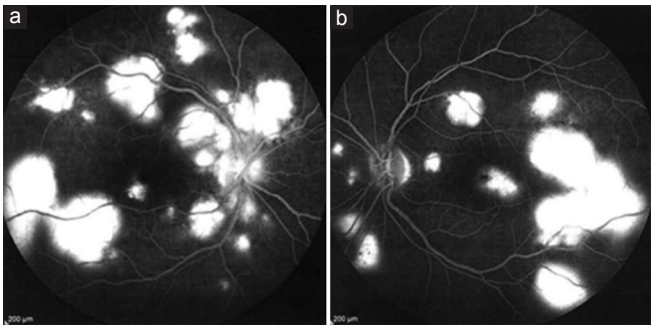

Figure 2. The later phases of fluorescein angiography of the right (2a) and the left eye (2b) at presentation, showing pooling of fluorescein in the subretinal spaces.

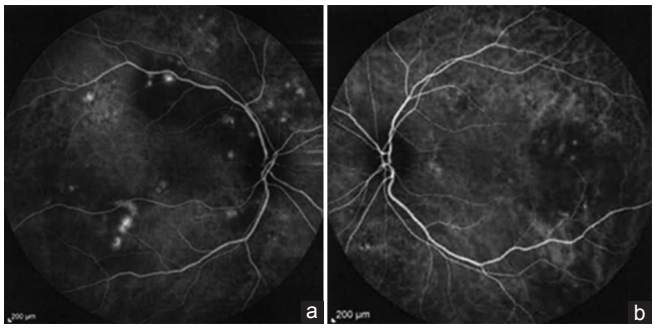

Figure 3. The early-phase indocyanine green angiography of the right (3a) and the left eye (3b) at presentation reveals dilated choroidal vessels and choroidal hyperpermeability.

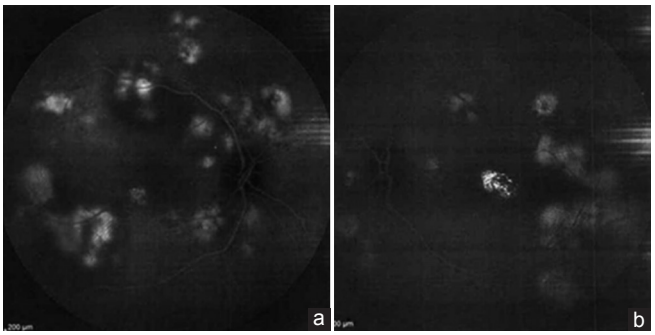

Figure 4. Later phases indocyanine green angiography of the right (4a) and the left eye (4b) showing choroidal hyperpermeability.

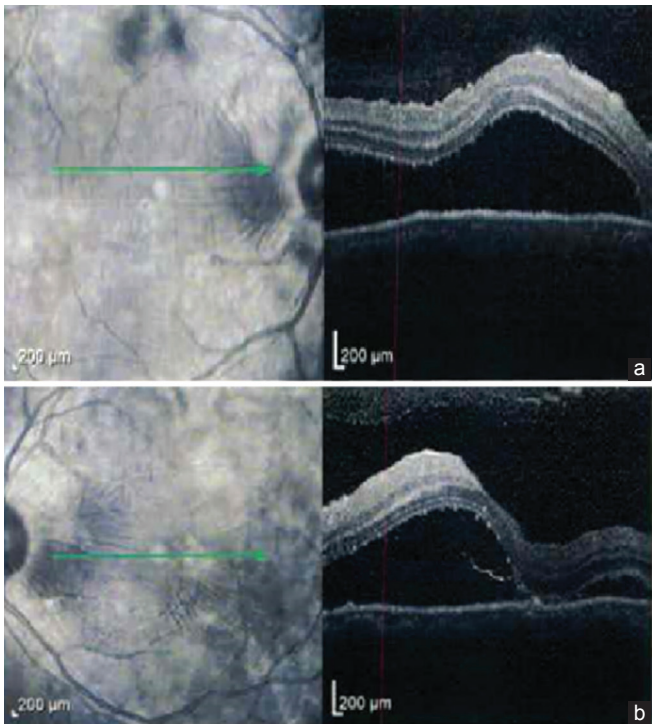

Figure 5. The infrared and optical coherence tomography images of the right (5a) and the left eye (5b) at presentation: accumulation of subretinal hyporeflective fluid with intact RPE layer is visible. The inner retina seems also to be edematous.

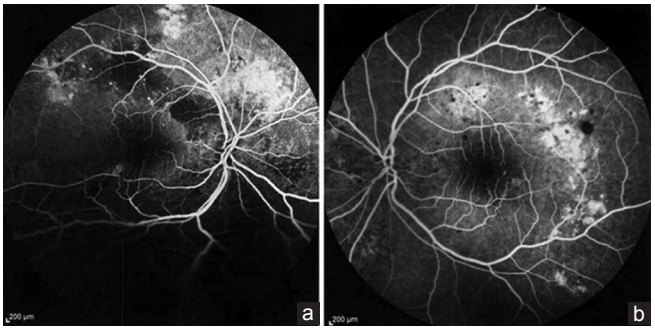

Figure 6. Fluorescein angiography of the right (6a) and the left eye (6b) four months after intravitreal bevacizumab injection: Leaking points have regressed significantly. Staining in favor of chronicity is seen bilaterally. While the subretinal fluid has decreased significantly in the right eye macula and left eye, but inferior subretinal fluis is steel present in the right eye.

angiopathy,[6] which results in choroidal ischemia, an important risk factor for development of CSCR, but in our case, the improvement of the clinical picture in spite of cyclosporine usage, makes this explanation less probable.

Increased level of systemic vascular endothelial growth factor (VEGF) is another possible etiologic factor, which sounds the most probable explanation in our case. Angiogenesis factors have a role in sustaining inflammatory responses seen following BMT,[7] and VEGF is a potent angiogenic factor whose role has also been shown in other chronic inflammatory diseases.[8] Although spontaneous reabsorption of subretinal fluid could occur in these cases,[4] the role of bevacizumab injection should be considered as an additional possibility. Bilateral response following unilateral intravitreal bevacizumab injection has been reported previously.[9] In the current case, the significant bilateral improvement following unilateral intravitreal bevacizumab injection supports the idea of systemic diffusion of bevacizumab following unilateral intravitreal injection.

This report shows that SRD following BMT might have a multifactorial pathogenesis. It also not only supports the possible role of increased systemic levels of VEGF in inducing bilateral multifocal SRDs following BMT, but also suggests the probable systemic diffusion of intravitreally injected bevacizumab, and its effect on the contralateral eye. Meanwhile, the fact that a single injection improved the situation for a long time could be attributed to the possible gradual remitting nature of the inflammation that probably occurs after BMT.

In conclusion, central serous retinal detachment is a possible complication following BMT, which probably has a multifactorial pathogenesis. One possible mechanism is increased levels of circulating VEGF, which may respond to intravitreal bevacizumab injection.

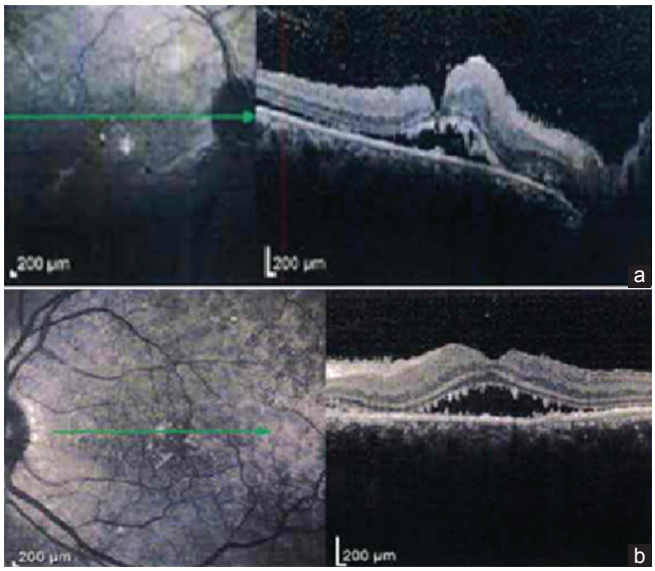

Figure 7. The infrared and optical coherence tomography (OCT) images of the right eye (7a) and the left eye (7b) four months after intravitreal bevacizumab injection: the subretinal fluid had decreased in both eyes.

Declaration of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of Interest

There are no conflicts of interest.

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