The Beneficial Effect of Hyperbaric Oxygen Therapy in A Case Having Cilioretinal Artery Occlusion Secondary to Familial Dysplasminogemia

Ailesel Displazminojemili Bir Olguda Gelişen Silyoretinal Arter Tıkanıklığı Sonrası Uygulanan Hiperbarik Oksijen Tedavisinin Yararı

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Case Report *Olgu Sunumu*

ABSTRACT

To report a case of unilateral cilioretinal artery associated with familial displasminogemia (FD) and its response to hyperbaric oxygen (HBO) treatment. A 34-year-old-man presented with acute loss of vision in the left eye because of the cilioretinal artery occlusion. Fundus photography, fluorescein angiography, OCT, visual field test were performed. HBO treatment was instituted to treat the patient who had been suffering for this complaint for a week. The patient had decreased plasminogen activity and elevated lipoprotein level and the diagnosis of FD was made by cardiologist. After the hyperbaric oxygen treatment, visual acuity improved from 20/200 to 20/25 and the size of scotoma in visual field test decreased from 30° to 20°.

The combined effect of decreased plasminogen activity and elevated lipoprotein (a) should be considered a possible cause of retinal artery occlusion. This case demonstrates a possible benefit of using hyperbaric oxygen treatment as a noninvasive tool in the treatment of cilioretinal artery occlusion.

Key Words: Cilioretinal artery occlusion, familial displasminogemia, hyperbaric oxygen therapy.

ÖZ

Bu çalışmada ailesel displazminojemili bir olguda gelişen tek taraflı silyoretinal arter tıkanıklığını ve hiperbarik oksijen tedavisine yanıtını bildirmeyi amaçladık. Otuz dört yaşında, sol gözünde silyoretinal arter tıkanıklığına bağlı gelişen akut görme kaybıyla başvuran erkek hastaya fundus resmi, flöresein anjiografi, optik koherens tomografi, görme alanı testi yapıldı. Bir haftadır yakınması devam eden hastaya hiperbarik oksijen tedavisi uygulandı. Hiperbarik oksijen tedavisinden sonra görme keskinliği 20/200'den 20/25'e yükseldi, görme alanındaki skotom 30°'den 20°'ye azaldı.

Azalmış plazminojen aktivitesi ve artmis lipoprotein (a) seviyesinin retinal arter ve ven tıkanıklıklarının olası sebebi olabileceği düşünülmelidir. Bu vaka ile silyoretinal arter tıkanıklığında noninvaziv bir yontem olan hiperbarik oksijen tedavisinin olası faydası gösterilmiştir.

Anahtar Kelimeler: Silyoretinal arter tıkanıklığı, ailesel displazminojemi, hiperbarik oksijen tedavisi.

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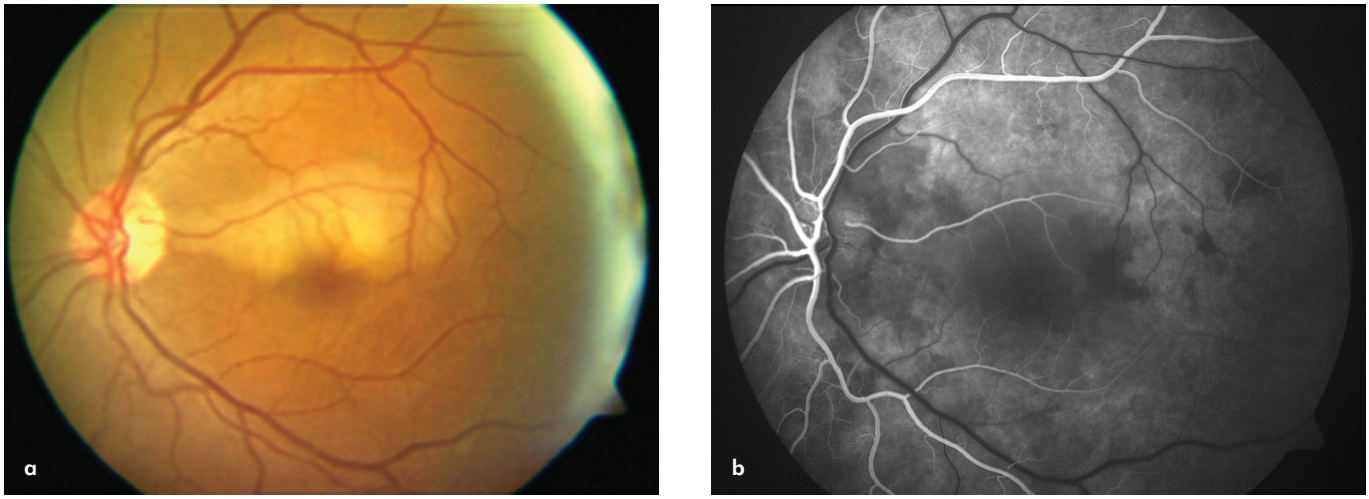

Figure 1: a) Fundus photograph showed white threadlike cil oretinal artery in the left eye. **b)** fluoresceine angiography showed delayed choroidal filling while having a normal cilioretinal artery filling in the left eye.

CASE REPORT

A healthy 34-year-old man had acute loss of vision in the left eye one week ago. He had a history of amaurosis fugax for one year. Ophthalmologic examination revealed the best-corrected visual acuity as 16/20 in the right eye (with correction of +5.75-3.00α140) and 20/200 in the left eye (with correction of +5.50-0.75α50). The axial length was 22.20 mm bilaterally. The anterior segment findings, pupils, and intraocular pressure were unremarkable. Fundus examination of the right eye was normal; the left eye had a white threadlike cilioretinal artery (Figure 1a). Fluorescein angiography showed delayed choroidal filling while having a normal cilioretinal artery filling in the left eye (Figure 1b). Automated perimetry revealed 30° scotoma adjacent to the fixation point in the left eye corresponding to ischemic areas (Figure 2a).

Serologic tests were negative including: anti-dsDNA, anti histon antibody, anti nukleozom, anti ribo p prot, Factor 5, ANA, CRP, RF, VDRL, cryoglobuline, anticardiolipin antibodies and lupus anticoagulant. Analyses for rheumatoid factor, complement factors C3 and C4, anti-neutrophil cytoplasmic antibodies with a cytoplasmic staining pattern (c-ANCA), and peripheral staining pattern (p-ANCA) were negative. Patient had a history of smoking 40 cigarettes a day for ten years.

Serologic tests was positive for lipoprotein a being 65.1 mg/dl (Normal range 1-30) and plasminogen activity was 47% (Normal range≥%70). Color Doppler images (CDI) of the left internal carotid artery disclosed total occlusion. His father's (52 years) and brother's (30 years) test results for lipoprotein were also high (62-54 mg/dl), without a history of a thrombotic event, they did not have plasminogen activity test results. Diagnosis of familial dysplasminogemia was established, and anticoagulant treatment with coumadin was initiated at the second week by internist.

After the ocular massage and parasenthesis , systemic asetozolamide and hyperbaric oxygen therapy was initiated immediately. 20 sessions of hyperbaric oxygen therapy lasting ten days was applied.The therapy was twice a day each session lasting 30 minutes and 240 kPa.The patient noted a decrease in the scotoma at the end of fourth session and automated perimetry showed a decrease in scotoma size to 20° from 30° (Figure 2b) at the end of first week.The visual acuity improved to 20/25 at the end of first month.

DISCOSION

Cilioretinal artery occlusion accounts for 5% of all retinal arterial occlusions. It can occur as an isolated event or in association with central retinal vein occlusion or less frequently anterior optic ischemic neuropathy.[1] When it occurs as an isolated vasoocclusive event, it has been related to embolic occlusion caused by atherosclerosis of the ipsilateral carotid artery, sickle cell trait and rheumatoid arthritis, oral contraceptives, high blood pressure.[2] Several studies indicated a relationship between familial dysplasminogenemia and, central retinal vein and cilioretinal artery occlusion,[3] macular choroidal occlusion,[4] and retinochoroidal circulatory disturbances,[5] although a single study showed no increased risk of thrombosis in heterozygous congenital dysplasminogenemia.[6] Hyperbaric oxygen therapy in retinal artery occlusion in addition to the standard therapy is used and seems to be beneficial but not certain.[7-8]

In general, visual field defects and fundus pallor can occur in areas that derive their blood supply exclusively from this vessel. A definitive diagnosis was made by fluorescein angiography, which shows a blockage or delay of blood flow in the cilioretinal artery. Occasionally, the occluded vessel is revascularized, hence producing normal findings in fluorescein angiography, which renders the diagnosis more difficult.

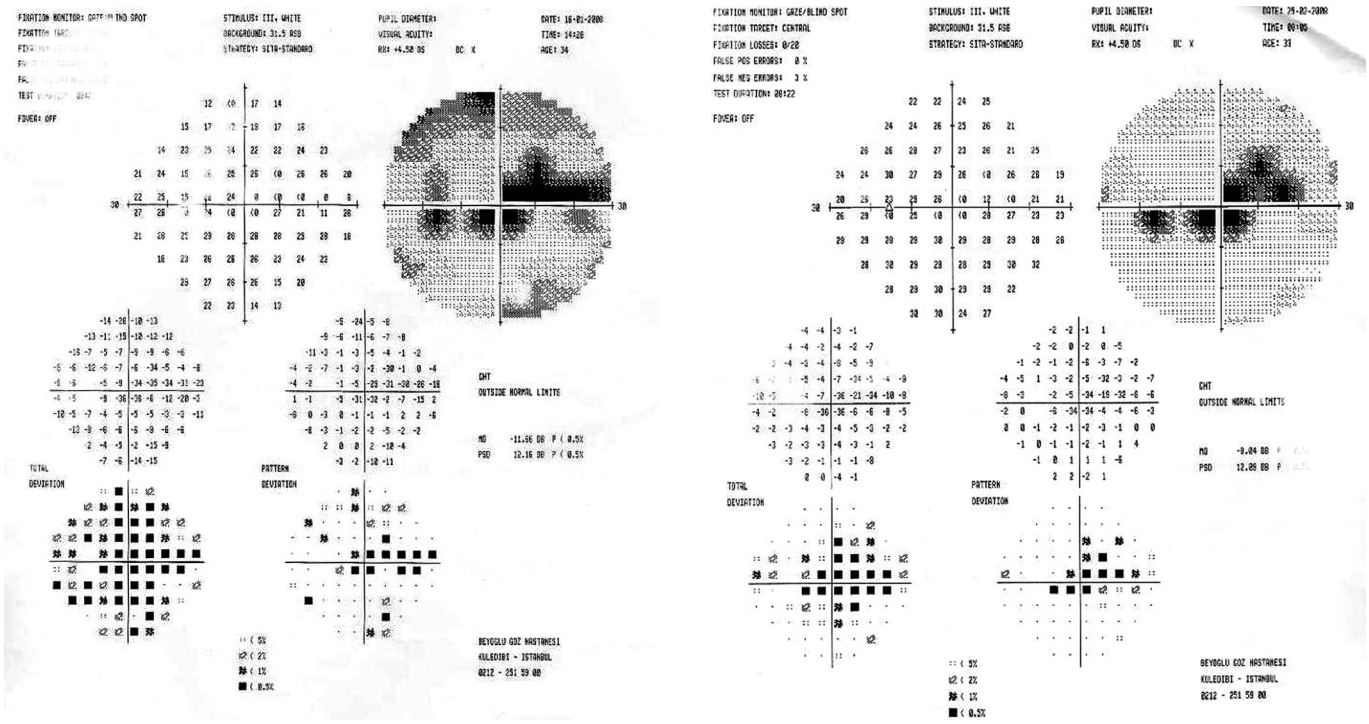

Figure 2: a) Visual field findings at first day : Automated perimetry revealed 30° scotoma adjacent to the fixation point in the left eye corresponding to ischemic areas. **b)** visual field findings at first week: Automated perimetry revealed 20° scotoma adjacent to the fixation point in the left eye.

But in this case, fundus pallor, visual field defects and OCT findings facilitated the diagnosis. No improvement was observed after ocular massage, parasenthesis and systemic therapy, and then hyperbaric oxygen therapy was initiated as a salvage treatment option. A decrease in scotoma size was noted after the fourth session by patient himself and automated perimetry showed a decrease in scotoma size to 20° from 30° (Figure 2b) at the end of first week and visual acuity improved to 20/25 at the end of first month. Visual acuity and visual field results also supported the subjective improvement. But the role of HBO therapy is still controversial in this case since the improvement could also have occurred spontaneously as well.

The correlation of familial displazminogemia with vascular thrombosis is unclear. The thrombotic tendency in this disorder may be responsible for the pathogenesis of cilioretinal artery occlusion. In our case, FD might have increased the tendency for thrombosis since no risk factor other than smoking was present. In young patients with retinal artery occlusion the incidence of blood component abnormalities is high. Therefore such blood disorders should to be sought in young and otherwise healthy patients with retinochoroidal circulatory disturbances.

In conclusion, in a young patient with retinal artery occlusion FD should be considered among the possible causes, a systematic evaluation for such hematological disorders should be carried out and hyperbaric oxygen therapy may kept in mind as an additional therapeutic modality.

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