Bilateral acute depigmentation of iris: 3-year follow-up of a case

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Abstract

Bilateral acute depigmentation of the iris (BADI) usually affecting young women, is a newly defined clinical diagnosis with bilateral symmetrical pigment loss of iris stroma without iris transillumination defect. Herein, we want to share the results of a 3-year-long follow-up of a 23-year old female patient with BADI. She was admitted to our clinic with a complaint of discoloration of both her brown irises. An ocular evaluation of the patient revealed symmetrical pigment deposition in trabecular meshwork. No iris transillumination defect, pupillary sphincter paralysis, keratic precipitates, and inflammatory reaction in anterior chamber were seen. The depigmented iris stroma became repigmented symmetrically after 3-year follow-up period. Although it is rare, BADI should be considered in the differential diagnosis of the diseases with bilateral iris depigmentation.

Keywords: bilateral acute depigmentation of iris, iridocyclitis, iris discoloration

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Introduction

Bilateral acute depigmentation of the iris (BADI), usually affecting young women, is a disease that has a favorable clinical course, with bilateral symmetrical pigment loss of iris stroma without iris transillumination defect.[1] The iris pigmentation can recover over time.[2] The etiology of BADI is not fully understood. The possibility of BADI should be kept in mind in the differential diagnosis of diseases of pigment spreading to the anterior chamber. This study was conducted in accordance with the tenets of Declaration of Helsinki. A written consent to publish the medical data and images was obtained from the patient.

Case report

A 23-year-old female patient was admitted to our clinic with a complaint of discoloration of both her brown irises. In her ophthalmologic examination, bilateral best corrected visual acuity was normal (10/10 with a standard Snellen scale), and intraocular pressure measured with Goldmann Applanation Tonometer was 13 mm Hg in both eyes. Symmetrical, grayish, and diffuse depigmentation with granular appearance was noticed in both iris stromas of the patient, except for 1- to 2-mm peripupillary band and 4- to 6-hour dial islands (Figure 1(a) and (b)). No iris transillumination defect, pupillary sphincter paralysis, keratic precipitates, and inflammatory reaction in anterior chamber were seen.

On gonioscopic examination, an intense pigment deposition was observed in the trabecular meshwork, while the angles were open in both eyes (Figure 2(a) and (b)). Dilated fundus examination was normal. Anterior segment optical coherence tomography images revealed normal iris configuration in both eyes (Figure 3(a) and (b)). The patient had no history of systemic disease. About 3 months before the onset of ocular symptoms, she had been hospitalized and treated with systemic antibiotics for pneumonia and urinary tract infection. She was treated for pneumonia with oral moxifloxacin for 2 weeks. Urinary tract infection was first treated with intravenous ceftriaxone but then it was replaced by ertapenem once beta-lactam (+) *Escherichia coli* was detected in the urine culture. She had no ocular complaints during her hospitalization and systemic treatment. After 3 months of discharge, iris discoloration was noticed by her friend by chance

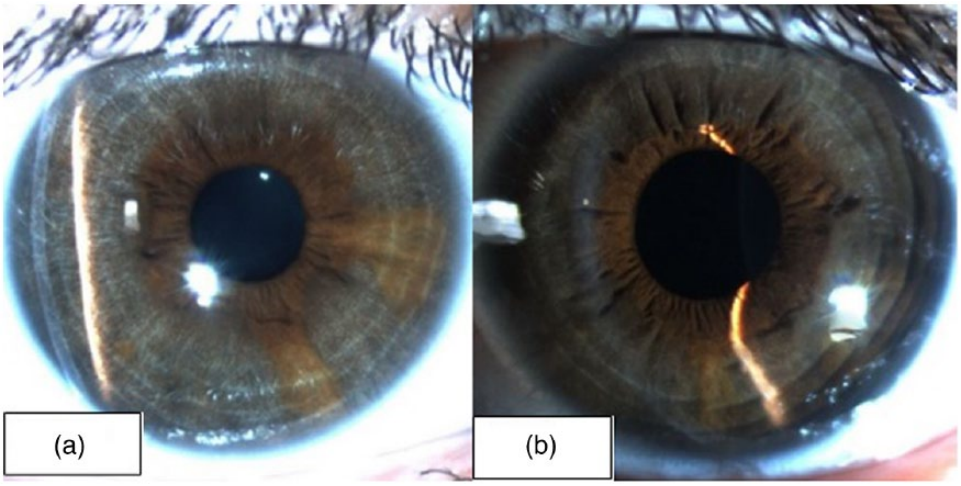

Figure 1. Color photographs of the right and left eye display symmetrical, grayish, and diffuse depigmentation with granular appearance, except for 1- to 2-mm peripupillary band and 4- to 6-hour dial islands: (a) right, (b) left.

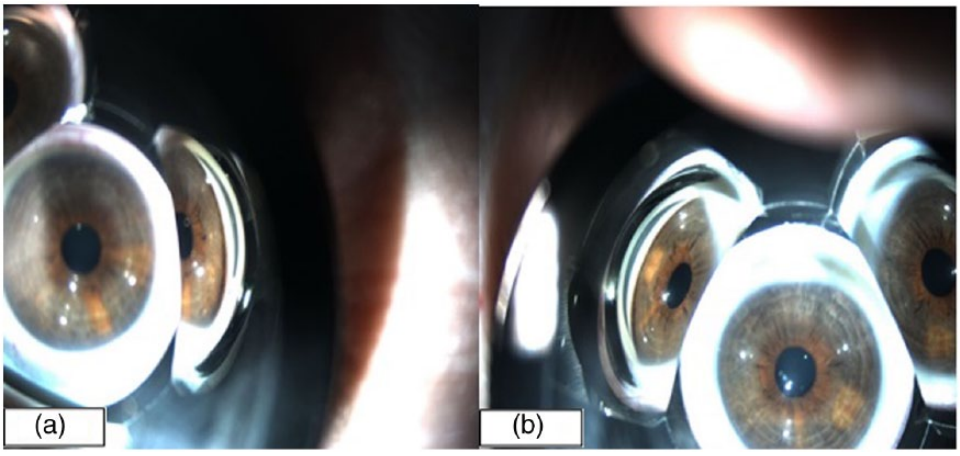

Figure 2. Gonioscopic color photographs of the right and left eyes display intense pigment deposition in the trabecular meshwork: (a) right and (b) left.

and thereupon she applied to our clinic. Routine laboratory work-up showed that her complete blood count, erythrocyte sedimentation rate, liver enzymes, blood urea nitrogen (BUN), creatinine, serum angiotensin-converting enzyme (ACE), C-reactive protein, and rheumatoid factor (RF) were all normal. Venereal Disease Research Laboratory (VDRL) and fluorescent treponemal antibody-absorption (FTA-ABS) tests for syphilis were negative. Serum immunoglobulin (Ig)-G and M antibodies against herpes simplex virus (HSV) 1 and 2, varicella zoster virus (VZV), cytomegalovirus (CMV), Epstein–Barr virus (EBV), rubella, and toxoplasma were normal except for high anti-rubella IgG and anti-CMV IgG. No complaints or complications were observed during the 3-year follow-up period. At the end of the 3 years, iris repigmentation was started in both eyes, although it was unable to reach predisease levels (Figure 4(a) and (b)).

Discussion

Bilateral acute iris depigmentation is a new definition first described in 2006 by Tugal-Tutkun and Urgancioglu.[1] BADI is a disease with good clinical course that usually affects young women and main finding of disease is symmetrical pigment loss in both iris stroma.[2] It is mistaken for the diseases caused by anterior chamber pigmentation such as Fuchs heterochromatic iridocyclitis, pigment dispersion syndrome, pseudoexfoliation glaucoma, and viral iridocyclitis due to HSV, VZV, and CMV.[2] Fuchs heterochromatic iridocyclitis differ from this disease only by the presence of bilateral involvement of 5–10% and the presence of satellite precipitates

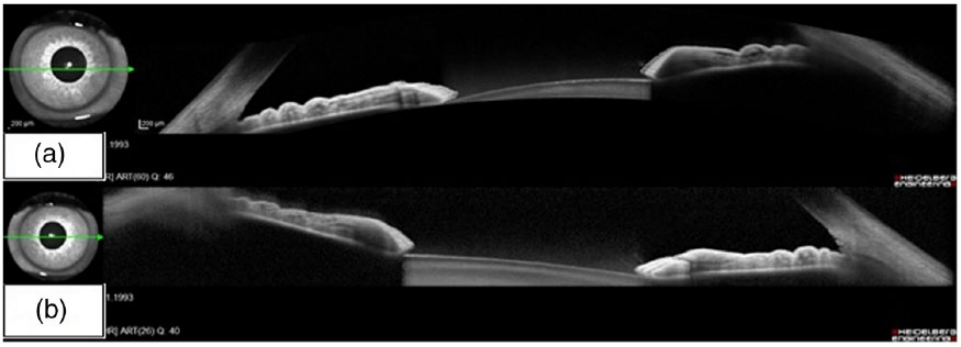

Figure 3. Optic coherence tomography of anterior segment. The right and left eyes show normal iris configuration: (a) right and (b) left.

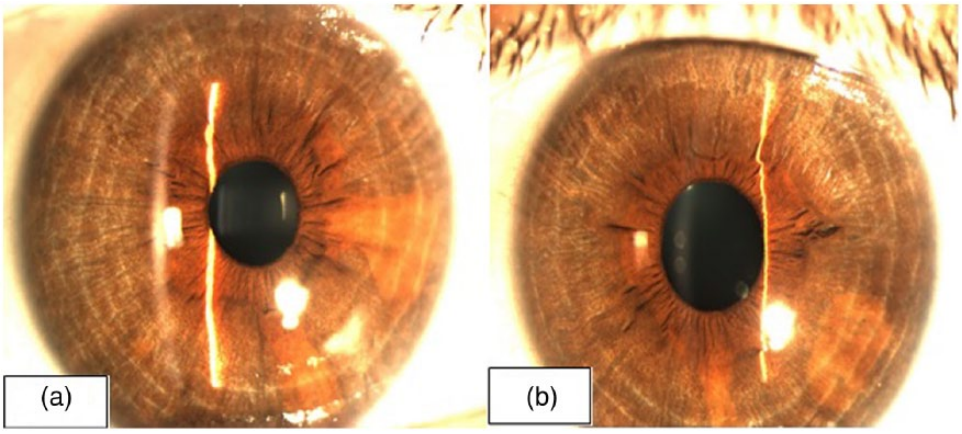

Figure 4. Color photographs of the right and left eyes after 3 years show iris repigmentation: (a) right and (b) left.

on the cornea.[3] Pigment dispersion and pseudoexfoliation syndromes are characterized by iris transillumination defects due to pigment discharge from the posterior pigment epithelium of iris and pigment accumulation not only in the trabecular meshwork but also in the zonules, and lens anterior capsules are different from BADI.[4] Herpetic iridocyclitis is a disease with unilateral acute anterior uveitis usually accompanied by decreased corneal sensation, increased intraocular pressure, iris transillumination defects, pupillary paralysis, and pupillary deformity which are the remarkable findings in the differential diagnosis of BADI.[5] Bilateral acute iris transillumination (BAIT), recently described by Tugal-Tutkun and colleagues[6] is also a rare disease that should be kept in mind in differential diagnosis of BADI.

The pigment stored in trabecular meshwork in BAIT is originated from the posterior pigment epithelium of iris and the most important findings are iris transillumination defect and sphincter paralysis. Otherwise, in BADI, iris stroma is the source of pigment spreading to the anterior chamber and deposited in the angle. Therefore, iris transillumination defect and sphincter paralysis do not appear in BADI, and iris discoloration with granular appearance may be reversible.[2,7] Often overlooked in practice and treated like iridocyclitis, diagnosis of BADI can be easily made with consideration paid to the examination findings clinically. The first one of these is stromal depigmentation and granular appearance of iris without iris transillumination defect, the second one is that there are no inflammatory cells in the anterior chamber and corneal endothelium even in acute attacks, and the third one is that it always displays bilateral symmetrical involvement.[2]

The etiopathogenesis of BADI is not fully known. The appearance of the first-defined case in summer gives rise to the thought of a different endemic virus or a different presentation of the herpetic eye disease.[1] However, in the study on 26 cases conducted by Tugal-Tutkun and colleagues,[2] no etiopathogenesis was observed in 42.3%, while 38.5% had the history of mild upper respiratory tract infection requiring such antibiotics as moxifloxacin and amoxicillin clavulanate. In fact, the

relationship between systemic moxifloxacin usage and BAIT has been emphasized in the studies.[8–10] However, BADI or BAIT is not seen in patients with the history of topical moxifloxacin usage, frequently prescribed in eye surgeries and well-penetration into eye. Because of this, the use of systemic moxifloxacin may not be the cause of BADI.[11]

Newly discovered resistant upper respiratory tract viruses may be cause disease in iris like conjunctiva. If so, whether it is BADI or BAIT in patient may vary, depending on the severity of the upper respiratory tract infection, the condition of the person's immune system, and the viral load. Our patient also had the history of systemic moxifloxacin, ceftriaxone, and ertapenem usage due to pneumonia and urinary tract infection during the summer of 2014.

The iris discoloration seen in BADI can return to normal in time. Two cases with BADI followed by Tugal-Tutkun and colleagues[2] were reported to have normalized iris after 4 years. In our case, the regaining of pigment in the iris was started at the end of the 3-year follow-up. The reversibility of iris depigmentation suggests that depigmentation may not be associated with active viral infection or inflammation of the iris tissue but may be due to a neuropathic effect.

BADI should be considered in the differential diagnosis of the diseases with bilateral iris depigmentation. In the majority of patients with BADI, pigment cells in the anterior chamber are considered to be inflammatory cells but they are misdiagnosed as iridocyclitis and exposed to the side effects of long-term topical corticosteroids use.

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Conflict of interest statement

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