histopathological feature of PBC and GCA is granulomatous inflammation. Thus, even though occurrence of GCA along with PBC and hypothyroidism is rare, awareness of this association may aid the management of this potentially blinding condition in elderly patients.

Competing interests: None

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DRS Anijeet[1], C Graham[2] and R Khooshabeh[1]

[1]Department of Ophthalmology, Stoke Mandeville Hospital, Aylesbury, Buckinghamshire, UK

[2]Department of Pathology, Stoke Mandeville Hospital, Aylesbury, Buckinghamshire, UK

Correspondence: R Khooshabeh, Department of Ophthalmology, Buckinghamshire NHS Trust, Stoke Mandeville and South Bucks Hospitals, Mandeville Road, Aylesbury, Buckinghamshire HP21 8AL, UK
Tel: +44 1296 315000;
Fax: +44 1296 315037.
E-mail: ramona.khooshabeh@sbucks.nhs.uk

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Sir,

Didanosine-induced retinopathy in adults can be reversible

Didanosine (DDI), a purine analogue used in the treatment of human immunodeficiency virus (HIV) disease, has been associated with pancreatitis, peripheral neuropathy, and retinopathy,[1,2] the latter being attributed to irreversible retinal pigment epithelium (RPE) loss accompanied by partial loss of the choriocapillaris and neurosensory retina in the mid-periphery.[3]

We present a case of a HIV patient with a clinical and electrophysiological diagnosis of DDI retinopathy (DDIR), whose electrophysiological abnormalities improved following DDI withdrawal.

Case report

A 53-year-old HIV positive Caucasian male presented with difficulty with his peripheral vision under scotopic conditions. His medical history included HIV disease, shingles, oral hairy leukoplakia, seborrheic dermatitis, and non-insulin-dependent diabetes with peripheral neuropathy but no diabetic retinopathy. At presentation, he had been HIV positive for 12 years with a $CD4^+$ T-cell count of 224 cells/μl and an undetectable viral load. He was on highly active antiretroviral treatment (HAART) with Didanosine, Tenofovir, Ritonavir, and Saquinavir, the Didanosine (400 mg od) for 5 years. Other medication included Gabapentin, Domperidone, Aciclovir, and Co-trimoxazole.

On examination visual acuity was 6/6 in each eye, with normal pupillary reactions. Anterior segment examination was unremarkable with normal intraocular pressures.

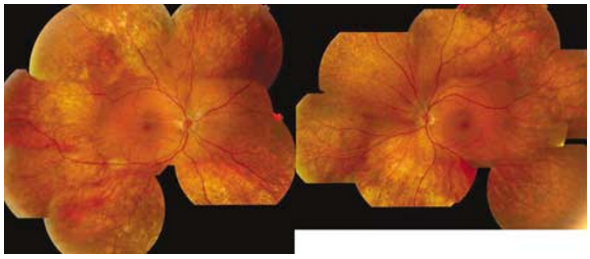

Figure 1 Fundus photographs of the right and left eye show extensive midperipheral atrophy at the level of the RPE in keeping with DDI retinopathy.

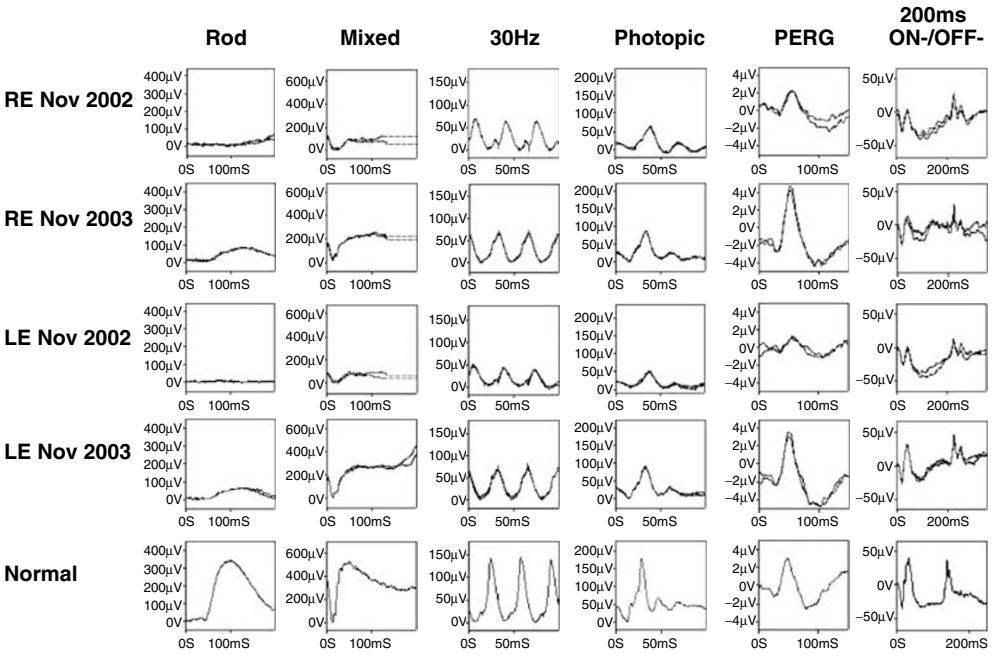

Figure 2 The ERG recordings from November 2002 and November 2003 demonstrate marked improvement in retinal and macular function following cessation of DDI. Note the improvement in rod ERG; the improvement in the a-wave of the mixed response confirming improvement at a photoreceptor level; both reduction in delay and improvement in amplitude in the cone-derived ERGs; and amplitude increase in the PERGs.

There were no inflammatory cells in the posterior chamber. Fundoscopy revealed bilateral midperipheral areas of retinal pallor surrounded by RPE clumping/stippling, but no signs of CMV retinitis, HIV retinopathy or vasculitis. The changes were consistent with DDIR. A 30-2 visual field and colour vision were normal. Visual electrophysiology demonstrated generalized retinal dysfunction with relatively severe rod involvement and marked cone system involvement (Figure 1). The left eye showed macular dysfunction. DDI was discontinued.

Repeat electrophysiology 1 year later revealed substantial improvement with only mild generalized retinal dysfunction (Figure 2). Macular function had normalized. The patient reported symptomatic improvement. The fundal appearances remained unchanged.

Comment

A case of DDIR is described of an adult who shows marked improvement in electrophysiological function following cessation of DDI. Similar improvement has previously only been described in a paediatric population.

Ocular complications resulting from DDI can include retinopathy[3–5] and optic neuritis.[4,6] DDIR was first reported in children,[4] and later in adults.[7] Retinal lesions, studied more extensively in children, appear as areas of RPE mottling and atrophy in the midperiphery, which later become circumscribed by RPE hypertrophy.[3,5] These lesions progress while the patient is on treatment but may stabilize after treatment is withdrawn.[3–5] Histopathological analysis in a child demonstrated multiple areas of RPE loss surrounded by areas of hypertrophy and hyperpigmentation of the RPE. Partial loss of the choriocapillaris and neurosensory retina were also noted.[3] Transmission electron microscopy demonstrated numerous membranous lamellar inclusions and cytoplasmic bodies in the RPE cells. These changes were seen in the retinal periphery, with the macula being spared both clinically and histologically. As hypopigmentation was noted histologically in a few perimacular RPE cells, Whitcup *et al* went on to postulate that 'it is possible that lesions of the macula or perimacular region may become apparent with prolonged didanosine use.' Clinically visible macular changes have not been reported as yet.

Electrophysiology in DDIR may reveal subnormal ERGs and a reduced EOG light rise.[3,4] Follow-up in one child demonstrated improved EOG light rise following withdrawal of DDI in association with stabilization of the lesions.[4]

Reversible retinal dysfunction, as noted in our case, could be related to reversal of RPE dysfunction.[4] The macula may make a full recovery due to the absence of areas of RPE atrophy and associated neuroretinal damage. However, due to the presence of these lesions in

the periphery, it may be that only a partial recovery is possible in this area. Alternatively didanosine may affect neuroretinal function, via an unknown mechanism independent of the RPE, which is reversed upon discontinuation of the drug.

In conclusion, a patient is described with visual symptoms and retinal lesions suggestive of DDIR. Electrophysiology revealed marked abnormalities of both rod and cone function that showed profound improvement following cessation of DDI. Similar findings have not previously been reported in an adult.

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AI Fernando[1], OA Anderson[1], GE Holder[2] and SM Mitchell[1]

[1]Department of Ophthalmology, Chelsea and Westminster Hospital, London, UK

[2]Department of Electrophysiology, Moorfields Eye Hospital, London, UK

Correspondence: SM Mitchell,
Department of Ophthalmology,
Chelsea and Westminster Hospital,
369 Fulham Road, London SW10 9NH, UK
Tel: +44 208 846 6131;
Fax: +44 208 846 6198.
E-mail: suzanne.mitchell@chelwest.nhs.uk

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Sir,

Oculodermal melanosis with choroidal melanoma in a black patient: a case report

Oculodermal melanosis (ODM) or Nevus of Ota is a congenital condition characterized by benign dermal melanosis of the skin in the area innervated by first, second, and rarely third division of the trigeminal nerve. Patients develop ipsilateral increase in pigmentation of the episclera, conjunctiva, uveal tract, and occasionally optic nerve head. In Caucasians, there is an association between ODM and uveal melanoma.[1,2] The disease, on the other hand, is rare in blacks. We report here a case of ODM and uveal melanoma in a black patient.

Case report

A 46-year-old Kuwaiti black female presented with a blind painful left eye of 1 year-duration. Patient is a known case of ODM involving the left side of her face. She had no light perception with intractable glaucoma despite full medical treatment. The right eye showed normal findings. The left eye revealed diffuse scleral pigmentation, corneal oedema, rubeosis iridis, and subtotal posterior synechiae (Figure 1a). The fundus could not be viewed because of vitreous haemorrhage. B-scan ultrasonography revealed a large choroidal mass in the posterior pole with total retinal detachment and vitreous opacities (Figure 1b). A-scan revealed high internal reflectivity of the mass suggestive of melanoma. Work-up for systemic metastasis was negative. Magnetic

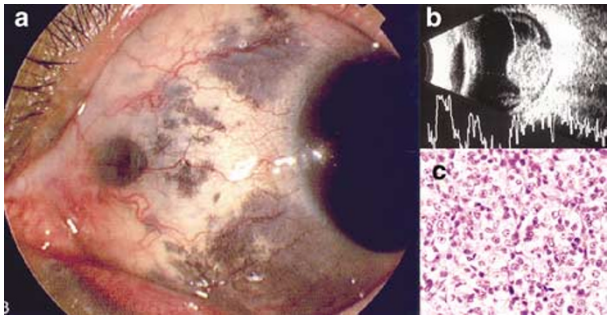

Figure 1 (a) Left eye scleral pigmentation. (b) B scan of left globe showing large mushroom choroidal elevation with retinal detachment. (c) The epithelioid melanoma cells showing large nuclei with prominent nucleoli (haematoxylin–eosin ×160).