In vivo confocal microscopic images of atypical amiodarone-induced keratopathy in patient with epithelial basement membrane dystrophy

Hidenori Inoue *, Koji Toriyama, Takeshi Joko, Atsushi Shiraishi

Department of Ophthalmology, Ehime University Graduate School of Medicine, Shitsukawa, Toon, Ehime, 791-0295, Japan

ABSTRACT

Keywords:
In vivo confocal microscopy
Amiodarone-induced keratopathy
Epithelial basement membrane dystrophy

A 73-year-old man was referred to the Ehime University Hospital with bilateral corneal opacities. His best-corrected visual acuity was 20/25 OU. The intraocular pressure was 13.0 mmHg and 18.0 mmHg in the right and left eyes. He was diabetic and had a myocardial infarction for which he was taking amiodarone 200 mg/day for 14 years. Slit-lamp biomicroscopy showed vortex-shaped opacities below the center of the cornea in both eyes which is typical of amiodarone-induced keratopathy (Fig. 1a and b). He also had oval-shaped opacities in the cornea of both eyes (Fig. 1c and d).

In vivo confocal microscopy (IVCM) with HRT-III RCM (Heidelberg Engineering, Germany) showed clusters of epithelial cells with reflective cytoplasm at the vortex of the opacities (Fig. 2a), and reflective microdots within the stroma. Similar findings have been reported in amiodarone-induced keratopathy. The IVCM also showed an abnormal epithelial basement membrane protruding into the epithelium as seen in patients with epithelial basement membrane dystrophy (EBMD; Fig. 2b). The IVCM-detected clusters of epithelial cells islands were detected in the ring-shaped structures extending from the basement membrane (Fig. 2c). They were co-localized with the oval-shaped opacities detected by slit-lamp microscopy. He had mild cataract and no abnormalities in the posterior segment of the eye. We performed regular medical examination annually, and the opacities were not treated. No significant changes were observed during these examinations.

1. Discussion

Amiodarone is a class III anti-arrhythmia drug with high affinity for lipids especially lysosomes, and their interactions result in intracytoplasmic lamellar inclusion bodies. These bodies are observed as vortex keratopathy by slit-lamp biomicroscopy, and they are seen as highly reflective epithelial cells by IVCM. Patients with EBMD are usually asymptomatic and may not be easily diagnosed by slit-lamp biomicroscopy because the findings vary. Because our patient did not have recurrent corneal erosions or slit-lamp biomicroscopic findings of EBMD, we could not make a diagnosis by slit-lamp alone. Thus, examination by IVCM showed the abnormal epithelial basement membrane protruding into the corneal epithelium, and the clusters of reflective corneal epithelial cells that were isolated into islands by the basement membrane. IVCM also detected microcysts in the epithelial cells, a characteristic finding of EBMD (Fig. 2d).

There have been no reports of the coexistence of these two diseases. Although this patient was asymptomatic, we could diagnose this rare condition by IVCM.

2. Conclusion

IVCM findings can provide evidence explaining the corneal morphological changes of vortex keratopathy and also the oval-shaped corneal opacities that were the amiodarone deposits in the epithelial cells.

Patient consent

Written consent to publish this case has not been obtained. This report does not contain any personal identifying information.

* Corresponding author. Tel.: +81 89 960 5361; fax: +81 89 960 5364.
E-mail address: inoue.hidenori.yo@ehime-u.ac.jp (H. Inoue).

https://doi.org/10.1016/j.ajo.2021.101105
Received 7 May 2020; Received in revised form 10 February 2021; Accepted 11 April 2021
Available online 2 May 2021
2451-9936/© 2021 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license
Fig. 1. Slit-lamp photographs of both eyes. A hole-like pattern of golden-brown deposits can be seen below the center of both corneas (a, right; b, left). An oval-shaped opacity can be seen in both corneas (c, right; d, left). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)
Funding

No funding or grant support.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The following authors have no financial disclosures: HI, KT, TJ, AS.

References

1. Ikegawa Y, Shiraishi A, et al. In vivo confocal microscopic observations of vortex keratopathy in patients with amiodarone-induced keratopathy and fabry disease. J Ophthalmol. 2018;2018, 5315137.
2. Labbe A, Nicola RD, et al. Epithelial basement membrane dystrophy: evaluation with the HRT II Rostock Cornea Module. Ophthalmology. 2006;113:1301–1308.

Acknowledgements

None.

Fig. 2. In vivo confocal microscopic findings of the right eye. 

a: Clusters of epithelial cells with highly reflective cytoplasm are present in the basal cell layer of the corneal epithelium. 
b: Abnormal epithelial basement membrane protruding into the corneal epithelium can been seen. c: Clusters of reflective corneal epithelial cells surrounded by ring-shaped structures. d: Microcysts within the corneal epithelium can been seen.