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Case Report

Bilateral Late-Onset Pigment Dispersion Syndrome following Implantable Collamer Lens Surgery: A Case Report

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Keywords
Pigment dispersion syndrome · Implantable collamer lens · Glaucoma

Abstract
Introduction: We report a case of bilateral pigment dispersion syndrome after 13 years of uncomplicated implantable collamer lens (ICL) surgery. ***Case Presentation:*** A 53-year-old woman was referred from her optometrist to our glaucoma clinic due to early superonasal visual field loss in both eyes. She was asymptomatic with no changes in visual acuity and had undergone bilateral ICL implantation 13 years ago to correct her high myopia. Clinical examination revealed pigment deposition on the corneal endothelium, iris transillumination defects, and iris vaulting at the areas of contact with the ICL. Gonioscopy showed open angles with significant pigmentation of the trabecular meshwork. The diagnosis of pigment dispersion syndrome secondary to ICL implantation was made, and subsequent follow-up visits demonstrated normal intraocular pressure IOP and stable visual fields. ***Conclusion:*** Pigmentary dispersion syndrome can occur several years after ICL implantation. This case report emphasizes the need for long-term follow-up and monitoring after ICL surgery.

Introduction

Phakic intraocular lenses (pIOLs) have emerged as a safe and effective surgical option for refractive error correction, offering an alternative to traditional laser vision correction procedures [1, 2]. The main advantages of pIOLs over photorefractive keratectomy and LASIK are that they can provide a broader range of treatable ametropia, faster visual recovery and a

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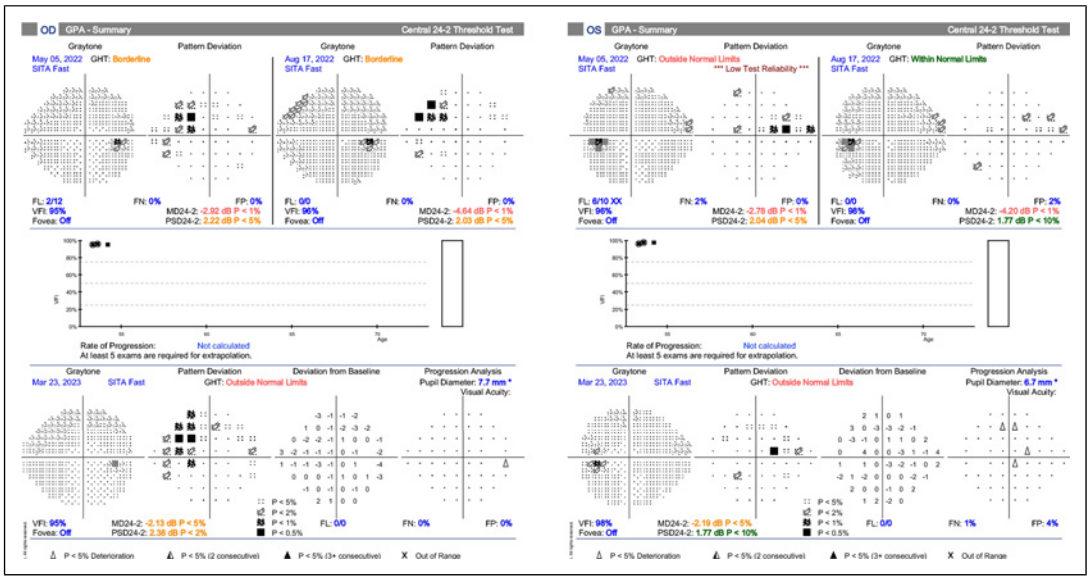

Fig. 1. Humphrey 24–2 VF testing of both eyes, showing early superonasal VF loss in both eyes across three examinations.

more stable refraction. Additionally, they offer the advantage of reversibility, as the pIOLs can be surgically removed, making the refractive result potentially reversible [1].

The Visian implantable collamer lens (ICL; STAAR Surgical, Monrovia, CA, USA) is currently the only FDA-approved posterior-chamber pIOL over the last 30 years. While ICLs have demonstrated excellent outcomes in terms of visual acuity and patient satisfaction, they are not without potential complications, which can be more disabling compared to the ones associated with keratorefractive surgeries [3]. These include anterior subcapsular cataract, pupillary block, and endothelial cell loss [3]. Pigment dispersion syndrome following ICL implantation has also been reported in a few case reports [4, 5], occurring usually after some weeks or months postoperatively, while pigmentary dispersion glaucoma has been described only in one study 8 years after ICL surgery [6].

This case report aimed to present a rare case of late-onset pigmentary dispersion syndrome 13 years after ICL implantation. To the best of our knowledge, this is the first documented case of pigmentary dispersion specifically associated with ICL implantation after such a long postoperative period.

Case Presentation

A 53-year-old woman was referred by her optometrist after a routine appointment to our glaucoma clinic (Department of Ophthalmology, Aberdeen Royal Infirmary, Aberdeen, UK) due to superonasal visual field (VF) loss in both eyes and family history of glaucoma. She was asymptomatic and has not experienced any change in her visual acuity during the last years. The patient had undergone uncomplicated bilateral ICL implantation (Visian ICMV4, −14 diopters, 13.2 mm) 13 years ago to manage her high myopia (−10 diopters in both eyes). Both her mother and grandmother had been diagnosed with open angle glaucoma in the past, while her past medical history was unremarkable.

Upon presentation, uncorrected visual acuity and IOP were 6/6 and 18 mm Hg, respectively, in both eyes. The central corneal thickness of her right eye and left eye (LE) was

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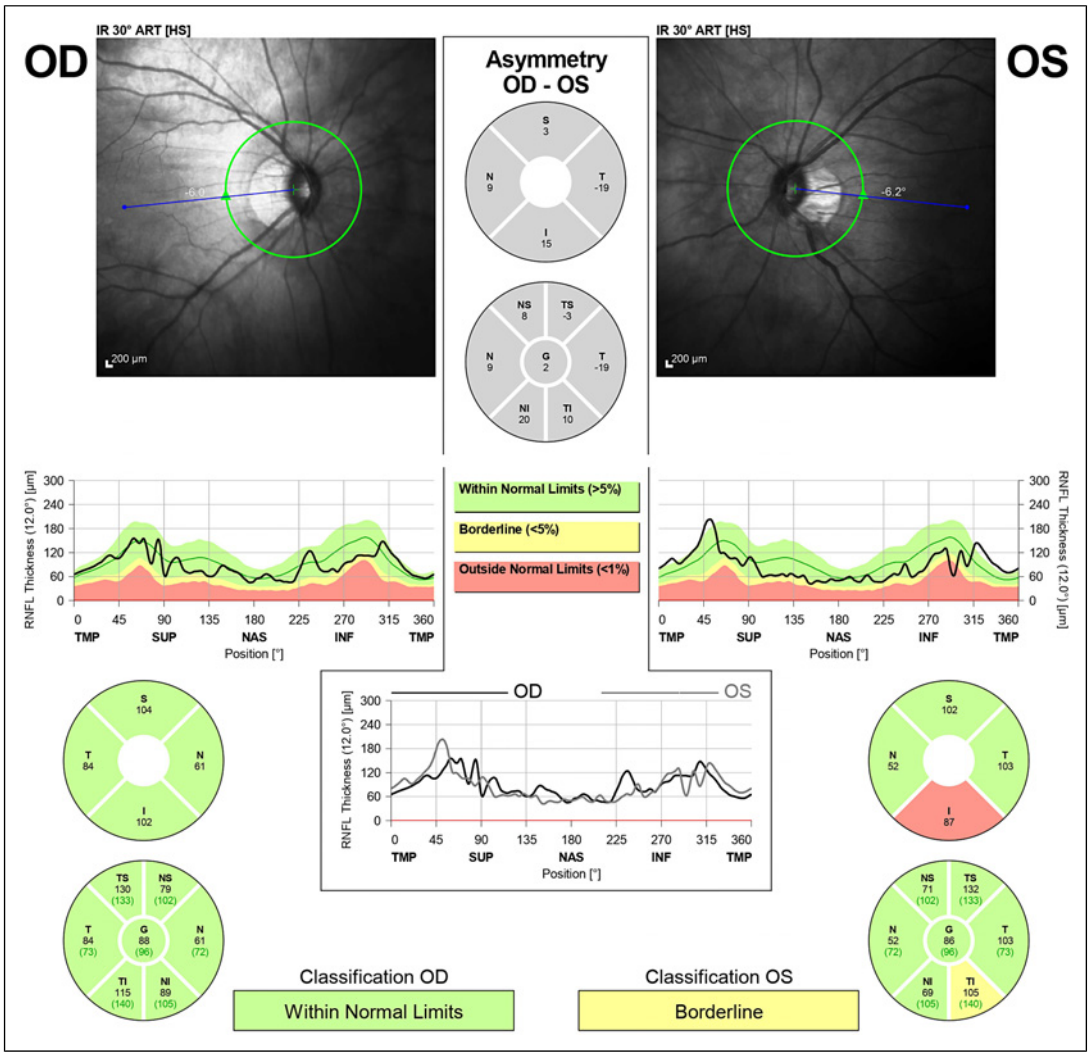

Fig. 2. Infrared imaging and retinal nerve fiber layer (RNFL) thickness analysis, showing highly tilted optic discs with peripapillary atrophy in both eyes and borderline RNFL decrease in the LE.

526 µm and 528 µm, respectively. VF testing with computerized automated perimetry (Humphrey 24–2 threshold white-on-white test, using the SITA-Fast algorithm with a size III stimulus) showed early superonasal VF loss in both eyes (right eye had a mean deviation of −2.92 dB with borderline glaucoma hemifield test, while the LE had a mean deviation of −2.78 dB with abnormal glaucoma hemifield test), with low test reliability of the LE due to high percentage of fixation loses (shown in Fig. 1). Retinal nerve fiber layer thickness measured by optical coherence tomography (Spectralis® OCT, Heidelberg Engineering, Heidelberg, Germany) was borderline decreased in the inferotemporal segment of the LE (shown in Fig. 2).

Slit-lamp biomicroscopy showed in both eyes pigment deposition on the corneal endothelium and on the anterior surface of the ICL, patent superior iridectomies, iris transillumination defects and iris vaulting at the areas of contact with the ICL (shown in Fig. 3, 4). These signs have not been present in her previous appointments at her community optometrist. Fundoscopy also revealed signs consistent with high myopia like tilted optic discs with peripapillary atrophy (shown in Fig. 2) without any glaucomatous optic nerve damage.

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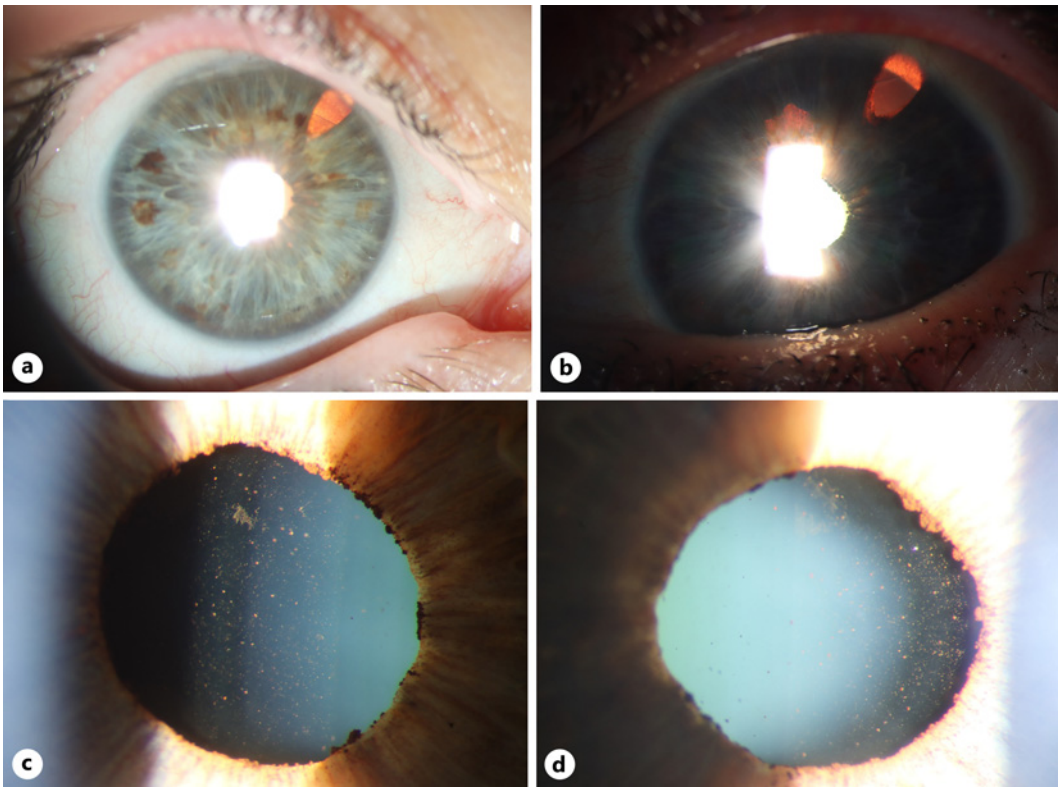

Fig. 3. Slit-lamp photography of the right eye (**a**) and LE (**b**), showing iris transillumination defects and the edge of the ICLs seen through the surgical iridectomy sites. Pigment deposition on the anterior surface of the ICL in the right eye (**c**) and LE (**d**) can also be seen.

Additionally, gonioscopy showed open angles bilaterally with grade 4+ pigmentation of the trabecular meshwork (shown in Fig. 4). Given these signs, the diagnosis of pigment dispersion syndrome secondary to ICL implantation was made, and we decided to monitor the patient closely. During the next visits (seven and 10 months after her first appointment) IOP was well controlled, while no changes were noted in the VF testing, optic nerve examination and pigment dispersion (shown in Fig. 1). The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000538547).

Discussion

In this report we described a case of late-onset pigmentary dispersion syndrome secondary to ICL implantation. The design of ICLs has been evolved during the last decades. Previous models of ICLs, including V0, V1, V2, and V3, had limitations due to a high incidence of cataract [7]. However, the introduction of the recent V4 model in 1996 significantly reduced the occurrence of cataracts. The V4 model incorporated additional vaults to minimize the risk of anterior subcapsular opacities, and it also featured four 360-μm holes to facilitate the removal of viscoelastic during surgery [1]. Finally, in the last model V4c a central 360-μm hole was added to improve aqueous humor flow and eliminate the need for peripheral iridotomies. The V4 and V4c models have demonstrated a significant decrease in reported complications,

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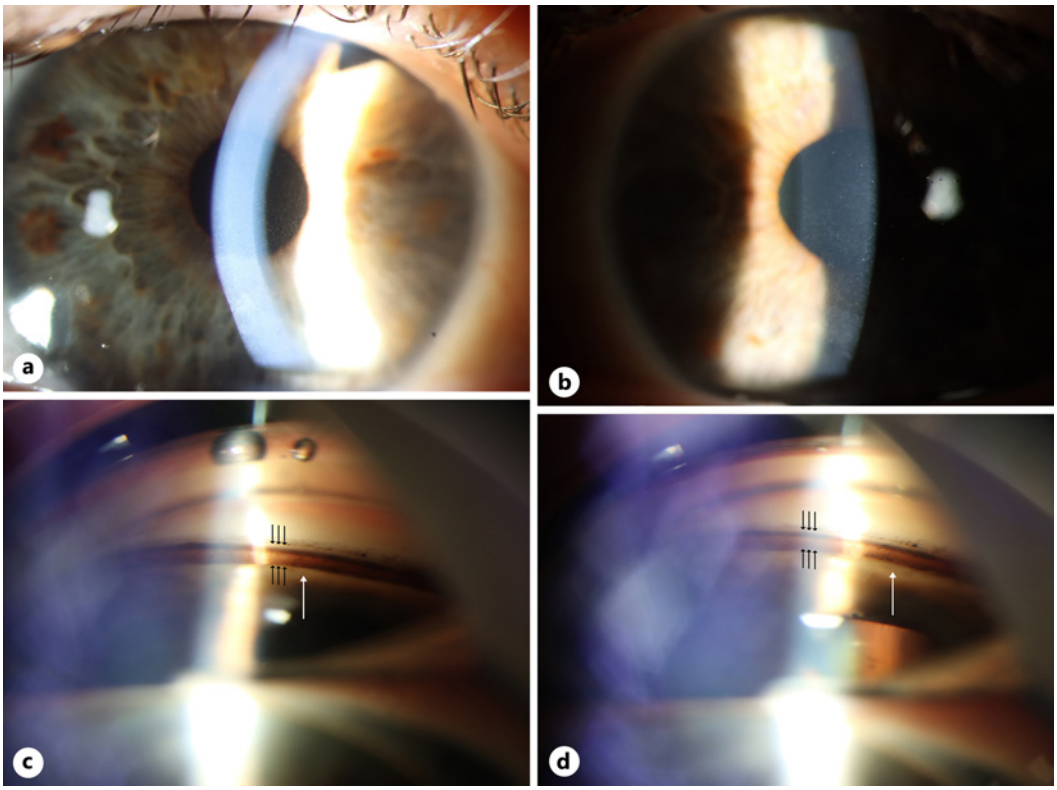

Fig. 4. Slit-lamp photography of the right eye (**a**) and LE (**b**), showing pigment deposition on the corneal endothelium on the anterior surface of the ICL. Grade 4+ pigmentation (black arrows) of the trabecular meshwork and iris forward vaulting at the areas of contact with the ICL (white arrows) seen in gonioscopy of the right eye (**c**) and LE (**d**).

probably due to the maintenance of normal anterior segment physiology, including aqueous humor flow through the central hole [7].

ICL implantation has also been found to affect iridocorneal angle structure and trabecular pigmentation, mainly during the early-postoperative periods [8]. Chung et al. [8] showed that both trabecular-iris angle and angle opening distance decreased significantly 1–3 months postoperatively, with no subsequent progressive changes. Trabecular pigmentation increased in 41.7% and 16.7% of patients during the early and late postoperative periods, respectively. However, stable IOP was observed in most cases, except for one eye which required anti-glaucoma medications. Additionally, in the manufacturer's product information leaflet [9] it is reported that during gonioscopy abnormal pigment suggestive of pigmentary dispersion and pigment on corneal endothelium was found in 6% and 1.2% of eyes ≥5 years postoperatively. Regarding the mechanism of this complication, it has been suggested that the implantation of the ICL may cause mechanical trauma to the iris, leading to the release of pigment granules into the aqueous humor [3]. Additionally, ICL may cause changes in the flow of aqueous humor, leading to increased deposition of pigment in the trabecular meshwork.

Patients who undergo ICL surgery are usually myopic, with a greater risk for open angle glaucoma, as well as pigment dispersion syndrome, compared to the general population. Given that ICL implantation can also be associated with pigment dispersion, myopic patients with ICL need close monitoring in order to identify early any glaucomatous changes. Only a few case reports have described the development of pigmentary dispersion syndrome with or without high IOP after the V3 model of ICL implantation. Sanchez-Galeana et al. [5]

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documented a case of a patient with pigmentary dispersion syndrome accompanied by a persistent elevation in IOP 6 months after the ICL implantation, which was treated with ICL explantation and trabeculectomy to lower the IOP. Similarly, in the study of Chung et al. [8] a case of elevated IOP in one eye was reported, characterized by a substantial increase in trabecular pigmentation just 1 week after the surgery, despite the ICL being positioned with a low vault. Notably, significant pigment deposits were observed on the surface of the ICL, necessitating the prolonged use of antiglaucoma medication. Finally, the only late-onset pigmentary glaucoma secondary to ICL implantation is described by Ye et al. [6], where a 50-year-old man developed pigmentary glaucoma 8 years postoperatively, and he was treated with removal of the ICL, cataract surgery and topical glaucoma medications.

Conclusion

Pigment dispersion syndrome secondary to ICL implantation can occur many years postoperatively. The aim of this report is to raise awareness of this atypical presentation and to highlight the importance of long-term follow-up and monitoring after ICL surgery. By sharing this case, we aim to contribute to the existing knowledge of the potential long-term complications associated with ICLs, specifically the development of pigmentary dispersion syndrome.

Statement of Ethics

Ethical approval is not required for this case report in accordance to national guidelines. A written informed consent form was obtained from the patient to publish these data and images.

Conflict of Interest Statement

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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Author Contributions

A.K. and M.K. contributed to the study conception and design, drafted and critically revised the manuscript for important intellectual content.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material files. Further inquiries can be directed to the corresponding author.

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