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A case of delayed recurrent hyphema following Implantable Collamer Lens (ICL) surgery

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ARTICLE INFO	ABSTRACT
<i>Keywords:</i> Recurrent hyphema UGH syndrome Intraocular lens iris chafing syndrome Toric ICL implantation	<i>Purpose</i> : To present a case of delayed recurrent hyphema following toric ICL implantation. <i>Observations</i> : This case reports a 24-year-old Asian female who presented with sudden decrease in vision in the right eye, accompanied by recurrent massive anterior chamber hemorrhage, six months after bilateral implantation of toric ICL with central holes for myopia correction. Despite initial conservative treatment with immobilization and intraocular pressure (IOP)-lowering medication at another hospital, the hyphema persisted. At our hospital, her corrected visual acuity (CDVA) in the right eye was counting fingers (CF) at 50 cm, with visible blood clots and hyphema in the anterior chamber. Initially, the patient was treated with a combination of three IOP-lowering medications: brimonidine eye drops, brinzolamide eye drops, and timolol eye drops, but the condition recurred. Two weeks later, we performed an anterior chamber hyphema evacuation and ICL removal surgery in the right eye. Postoperatively, the patient's IOP stabilized and her vision gradually recovered. One month after the surgery, a follow-up examination showed a CDVA of LogMAR 0.6 in the affected eye.

Conclusion and importance: This case report is essential for characterizing a rare and serious complication following toric ICL implantation, highlighting the importance of close monitoring and timely intervention.

1. Introduction

In recent years, the prevalence of myopia has significantly increased across all age groups.^{1–3} With the improvement in quality of life, the demand for refractive surgery has also markedly risen. Among these, Implantable Collamer Lens (ICL) surgery is a well-established ophthalmic procedure commonly used to correct myopia, hyperopia, or astigmatism. This surgery involves implanting a soft, biocompatible artificial lens, often made of materials such as CollamerTM, a collagen copolymer that combines acrylic and collagen, or silicone, into the eye.⁴ This provides an effective treatment option for patients with corneas unsuitable for traditional laser surgery or those with high refractive errors.⁵ ICL/TICL is composed of a collagen called collagen protein,

which is not affected by the eye's immune system.⁶ The first-generation ICL/TICL had various defects, such as poor predictability due to lens design issues. For the second-generation (V2) and third-generation (V3) models, the incidence of pupillary block glaucoma and pigment dispersion glaucoma has decreased.⁷ Although ICL surgery typically yields good outcomes and rapid recovery, complications may occur in some patients. These include collamer lens dislocation,^{8,9} anterior subcapsular cataract, reverse orientation of the ICL, glucoma,¹⁰ glare, and halos.¹¹ Despite numerous studies on ICL surgery and its complications, reports of delayed recurrent hyphema are exceedingly rare. After conducting a literature review on February 1, 2024, utilizing PubMed and Google Scholar with the keywords "ICL complications," "UGH Syndrome," and "Hyphema," we found limited reports on delayed recurrent

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hyphema following ICL surgery. This case report aims to present a unique instance of this complication, contributing valuable insights to the existing literature and clinical practices.

Delayed hyphema refers to hyphema occurring at least two months after ocular surgery or trauma.¹² Its impact on vision and treatment can cause varying degrees of distress for patients. Common causes of delayed hyphema include increased external eyelid venous pressure, individual factors, surgical factors, prolonged sleeping on the operative side, and position-related intraocular pressure reduction.¹³ Currently, reports of late-onset recurrent hemorrhage following ICL surgery are rare. Our search revealed only one published case report in April this year on UGH syndrome following implantation of the older model ICL. Thus, ongoing discussion and attention to recurrent anterior chamber inflammation after ICL surgery are crucial for providing robust analytical support in future research.

This case aims to explore a case report of delayed recurrent hyphema following ICL surgery, focusing on its etiology and diagnosis. We delved into the potential mechanisms, risk factors, clinical manifestations, and treatment strategies. By elucidating the relationship between these factors, we hope to enhance the understanding of rare complications following ICL surgery and optimize treatment protocols for affected patients.

2. Case report

A 24-year-old Asian female underwent toric ICL implantation (model VTICMO12.1; Toric Myopic 12.1mm; Collamer[™]) in both eyes at an external hospital six months prior. Her preoperative measurements indicated a right eye anterior chamber depth of 2.94 mm and a white-towhite (WTW) distance of 10.8 mm. Additionally, her preoperative keratometry values were K1: 44.69@160 and K2: 48.06@70. On the first day post-ICL implantation, her vault measurements were 626 µm in the right eye and 608 µm in the left eye. One month postoperatively, the vault measurements were 463 μm in the right eye and 470 μm in the left eye. She presented with a chief complaint of sudden vision loss in her right eye, triggered by tilting her head forward while washing her hair. Her medical and systemic history were unremarkable. Examination revealed recurrent anterior uveitis, significant hyphema, and elevated intraocular pressure (IOP) in the right eye. The patient had experienced mild blurry vision a few days before the acute vision decline. Prior to presenting at our hospital, she had undergone two weeks of conservative treatment involving immobilization and IOP-lowering medication, but recurrent hyphema persisted.

At our hospital, her corrected visual acuity (CDVA) in the right eye was counting fingers (CF) at 50 cm. A blood clot was visible in the pupil area of the anterior chamber, with approximately 3 mm of hyphema

inferiorly (Fig. 1). The posterior segment was not clearly visible, and the IOP was 40 mmHg. Ancillary tests included ultrasound biomicroscopy (UBM), which indicated significant hyphema in the anterior chamber (Fig. 4), and B-scan ultrasonography, which ruled out suprachoroidal hemorrhage. Anterior segment optical coherence tomography (AS-OCT) excluded excessive vaulting of the ICL. Conjunctival vessel optical coherence tomography angiography (OCTA) showed no significant vascular abnormalities. The intraocular pressure of the left eye was 16 mmHg, with an uncorrected visual acuity of 1.0. No significant abnormalities were observed in the anterior chamber, as shown in Figs. 5, 8 and 9.

Initially, the patient received a combination of three IOP-lowering medications; however, the anterior chamber inflammation in the right eye persisted, and the IOP fluctuated between 28 and 50 mmHg. Two weeks later, the patient underwent an anterior chamber hyphema evacuation and ICL removal surgery in the right eye. During the surgery, after removing the blood clots adhered to the surface of the ICL, a significant amount of fresh blood was observed gushing out from the central hole of the ICL in a jet-like manner (Figs. 6 and 7), particularly when the IOP was lower, although the exact source of the bleeding was not identified. Postoperatively, the patient's IOP stabilized within the normal range. On the first postoperative day, the corrected visual acuity (CDVA) in the right eye was 0.2; on the second day, it was LogMAR 0.1 (Fig. 2); and on the third day, it was LogMAR 0.6-. The patient was subsequently discharged. Two weeks later, a follow-up examination showed a CDVA of LogMAR 0.7 in the right eye (Fig. 3).

3. Discussion

Complications associated with ICL are rare and may include early or late anterior subcapsular cataract, elevated intraocular pressure, angleclosure glaucoma, iris transillumination defects, retinal detachment, and corneal endothelial cell loss.¹⁴ However, in this case, the patient's right eye experienced recurrent significant active hyphema six months after ICL implantation. Therefore, the etiology and subsequent diagnosis of this condition warrant further investigation.

One possible cause is that intraocular implants, such as Implantable Collamer Lenses (ICL) or Intraocular Lenses (IOL), may lead to recurrent hyphema due to improper positioning or mechanical friction with intraocular structures. This friction mainly results from the movement of the implant, especially after blunt trauma to the eye, ^{9,15} which can cause lens displacement and ocular damage. The resulting local inflammation, bleeding, and elevated intraocular pressure can further lead to hyphema and decreased vision. Although the ICL is designed to remain stable within the eye, the risk of mechanical friction persists if the implant is positioned incorrectly or the patient's ocular anatomy is incompatible.



Fig. 1. A blood clot was visible in the pupil area of the anterior chamber, with approximately 3 mm of hyphema observed in the inferior region.



Fig. 2. The hyphema in the anterior chamber was cleared on the first day after ICL removal surgery, and no signs of bleeding were observed.



Fig. 3. Two weeks after ICL removal surgery, follow-up examination showed no inflammatory reaction in the anterior chamber, and the pupil was slightly dilated.



Fig. 6. During the surgery, after removing the blood clots adhered to the surface of the ICL, a significant amount of fresh blood was observed gushing out in a jet-like manner from the central hole of the ICL.



Fig. 4. Ultrasound biomicroscopy (UBM) indicated significant hyphema in the anterior chamber.



Fig. 5. The postoperative UBM results showed no significant abnormalities in the structure of the ciliary body in the right eye.

enjamin Zhou and his team reported a case of UGH syndrome occurring two years after the implantation of a toric IOL during cataract surgery. This report suggests that UGH may be due to the toric IOL haptic being improperly placed outside the capsular bag during initial implantation or due to iris chafing when adjusting or rotating the IOL to align the toric axis. Another hypothesis is that a larger capsular bag diameter could lead to haptic dislocation during rotation, with the tilted IOL causing friction between the anterior haptic and the posterior iris, leading to anterior chamber inflammation and hyphema.¹⁶ In our case, the patient had a toric ICL implanted six months prior, and no significant axial movement of the lens was observed, ruling out the hypothesis of gradual ICL rotation causing iris chafing damage. Although literature on hyphema induced by ICL surgery is relatively scarce, the patient's

Fig. 7. During the surgery, after removing the blood clots adhered to the surface of the ICL, a significant amount of fresh blood was observed gushing out in a jet-like manner from the central hole of the ICL.

Fig. 8. No significant abnormalities were observed in the anterior chamber of the left eye, indicating a normal postoperative condition.

symptoms—significant and recurrent hyphema in the right eye—suggest a possible association with the initial ICL implantation. Given the absence of significant inflammatory response, mechanical factors such as friction between the ICL and iris or potential microtrauma during the initial surgery are more likely causes of the recurrent bleeding. Despite

Fig. 9. No significant abnormalities were observed in the anterior chamber of the left eye, indicating a normal postoperative condition.

being unable to clearly identify the specific bleeding source during surgery (possibly from the posterior iris surface or the ciliary body), the observation of fresh blood jetting from the central hole of the ICL after removing blood clots on its surface suggests possible mechanical damage and vascular rupture. Despite these findings, the possibility of small vascular anomalies or arterioles rubbing against certain portions of the ICL in unscreened ocular quadrants cannot be entirely ruled out. These could potentially cause hyphema bleeding, even though they were not identified using our OCT system.

The second hypothesis is that sudden positional changes triggered the anterior chamber hemorrhage. In the existing literature, positioninduced anterior chamber inflammation and other complications are more common in cases with IOL implantation due to zonular dysfunction. Zonular dysfunction, often caused by pseudoexfoliation, can lead to IOL instability and movement with positional changes, irritating the uveal tissue.¹⁷ Danny et al. reported a case of recurrent UGH syndrome after IOL implantation, triggered by yoga practice, especially involving prolonged prone positions. Clinical confirmation of IOL movement during positional changes was made using supine and prone UBM examinations.¹⁸ In our case, the patient experienced sudden vision loss after bending over to wash her hair, slight vision blurring upon straightening up, and significant vision decline two days later. Unfortunately, our hospital currently lacks the equipment to compare UBM images of the ICL and ciliary body in supine and prone positions. Furthermore, although a large amount of blood was observed jetting from the central hole of the ICL during surgery, no obvious blood stains were found on the anterior surface of the removed lens. Therefore, we hypothesize that the patient's zonular function may not have been significantly compromised, but we cannot entirely rule out the possibility that sudden positional changes caused a transient displacement of ocular structures, resulting in trauma to the vascular structures of the iris or ciliary body. This trauma likely led to fresh bleeding and subsequent intermittent hemorrhage.

The third possible explanation involves vascular anomalies within the eye. Although there is no direct evidence of vascular anomalies in our case, sudden changes in head position may exacerbate the position of the ICL concerning a potential vascular anomaly, leading to significant bleeding from either the ciliary body or the posterior iris. The rationale for this mechanism is that minor movements of ocular structures could exert shear force or pressure on fragile blood vessels, causing hemorrhage. While we currently lack direct imaging evidence to support the hypothesis of vascular anomalies, this possibility cannot be ignored.

A differential diagnosis to consider is UGH syndrome (Uveitis-Glaucoma-Hyphema syndrome), a rare but serious complication characterized by chronic recurrent anterior chamber bleeding, persistent uveitis, and secondary glaucoma.¹⁹ This syndrome is typically reported

after the implantation of anterior chamber lenses or improperly positioned posterior chamber lenses (IOL).^{20–22} This year, Khaled G. Almasri et al. reported the first case study of UGH syndrome following the implantation of non-centered perforated Visian ICL.²³ The patient developed asymmetrical anterior chamber inflammation two years after bilateral ICL implantation, with the left eve being more severely affected. Ancillary examination revealed pupil decentration in the left eve and multiple cysts in the scleral bag on UBM. Analysis indicated that temporal scleral cysts influenced the ICL position, exerting pressure on the ciliary body, causing iris chafing, and resulting in UGH syndrome. This case represents the first reported incidence of UGH syndrome following ICL surgery. Unlike our case, this patient had non-centered perforated ICLs implanted ten years prior, which restricted communication between the anterior and posterior chambers, unlike the more commonly used centrally perforated ICLs today. Additionally, the older ICL surgical process was more complex and time-consuming, increasing the risk of relying on peripheral iridectomy and potential ocular trauma. Finally, ancillary examinations revealed scleral cysts in the left eye, leading to ICL displacement and subsequent UGH syndrome, providing a more definitive etiology and diagnosis. In our case, the patient primarily presented with recurrent, significant hyphema in the right eve, which may have masked the underlying condition due to the significant anterior chamber bleeding, and multiple ancillary examinations, particularly UBM, did not reveal structural abnormalities within the eye. The high vault measurement of the ICL was 463 µm, within the normal range of 250-750 µm.

4. Summary

In this case report, we present a delayed and recurrent anterior chamber hemorrhage following ICL surgery and outline a feasible treatment approach. The patient experienced recurrent hyphema in the right eye six months after bilateral toric ICL implantation. Initial treatments were ineffective, but the condition was ultimately resolved through the removal of the anterior chamber hemorrhage and the ICL. Although the exact etiology remains unclear, we propose several possible diagnoses, including mechanical friction between the intraocular implant and intraocular structures, intraocular vascular anomalies, and position-induced recurrent anterior chamber hemorrhage. The patient's condition might also result from a combination of these factors. This report aims to alert ophthalmologists to the potential rare complications following ICL surgery and to provide insights into effective diagnostic and management strategies. Further research and case accumulation will help us better understand and manage similar cases, ultimately improving patients' quality of life.

Patient consent

Written informed consent was obtained from patients for publication of these case reports and any accompanying images.

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Authorship

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

CRediT authorship contribution statement

Hangjia Zuo: Writing – original draft, Methodology, Data curation, Conceptualization. Yonglin Chen: Data curation. Meiting Lin: Data curation. Hong Chen: Data curation. Shijie Zheng: Data curation. Wenjuan Wan: Writing – review & editing, Data curation, Conceptualization. Ke Hu: Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Data curation, Conceptualization.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ajoc.2024.102158.

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