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Case Report

A Rare Case of Lens-Induced Uveitis with Proliferative Vitreoretinopathy and Massive Encapsulation of Fallen Nuclear Material

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Keywords
Cataract surgery · Uveitis · Lens-induced uveitis · Proliferative vitreoretinopathy · Vitrectomy

Abstract
We experienced a rare case of lens-induced uveitis (LIU) with severe proliferative vitreoretinopathy (PVR) diagnosed upon finding lens nuclear material encapsulated by intravitreal proliferative tissue. A 60-year-old man was referred to our hospital for the treatment of vision loss caused by unexplained uveitis in the right eye (OD). Seven months previously, a complicated cataract surgery that required unplanned anterior vitrectomy and transscleral suture of intraocular lens was performed on that eye at another clinic. Severe inflammation with dense vitreous opacity occurred in the OD postoperatively. Although topical and oral administration of steroids reduced the inflammation 7 months after the surgery, PVR with tractional retinal detachment was developed in the OD. Pars plana vitrectomy (PPV) was performed for the treatment and diagnosis. PPV revealed the presence of lens nuclear fragments within the vitreous, which was approximately 60% the ordinary nucleus size and was encapsulated by intravitreal proliferative tissue. The nuclear fragments were extracted from a superior corneoscleral flap. Intraocular inflammation was reduced with postoperative topical and oral steroid treatments and the retina remained reattached 1 year after the PPV. In conclusion, uveitis with an episode of a complicated cataract surgery may suggest LIU.
Introduction

Lens-induced uveitis (LIU) is an inflammatory response to lens fragments caused by the breakdown of anterior chamber-associated immune deviation [1]. Due to the variety of potential causes (surgery, trauma, spontaneous lens capsule rupture, etc.), LIU presents with various ocular findings [1]. Retained lens fragments in cataract surgery are reported to cause LIU in up to 20% of cases [2]. Reported causes include not only dislocation of the lens nucleus into the vitreous cavity associated with posterior capsule rupture [3], but also retention of nuclear fragments in the anterior chamber angle after successful completion of surgery [4]. Although medical treatments including local and/or systemic administration of steroid are usually effective, lens fragments must be removed surgically when LIU is resistant to medical treatment [1, 5].

Here we report a case of treatment-resistant LIU with severe proliferative vitreoretinopathy (PVR) diagnosed at the time of vitrectomy, which found the lens nucleus encapsulated by intravitreal proliferative tissue.

Case Report

A 60-year-old man was referred to our hospital for treatment of vision loss caused by unexplained uveitis in the right eye (OD). He had no other notable systemic illness or symptoms except that he had atopic dermatitis. Seven months previously, a cataract surgery for hypermature cataract with complications that required unplanned anterior vitrectomy and transscleral suture of intraocular lens was performed in the OD at another clinic. Three months after the surgery, panuveitis with severe vitreous opacity, which made fundus observation difficult, developed in the OD, and was treated with both topical and oral administration of steroids. Seven months after the surgery, fundus observation became possible, but PVR with tractional retinal detachment was found to have developed in the OD. The patient was then referred to our hospital for the diagnosis and treatment of this unexplained panuveitis with PVR.

At the initial visit to our hospital, the Landolt decimal best corrected visual acuity (BCVA) was 0.01 in the OD, and the intraocular pressure was 7 mm Hg OD. The slit lamp microscopic findings in the anterior segment revealed exacerbation of inflammation accompanied with hypopyon (Fig. 1a). Optical coherence tomography revealed tractional retinal detachment and extension of the proliferative membrane to the macula (Fig. 1b). Blood examination revealed no specific findings related to ocular inflammation except HLA typing was positive for A26. Subconjunctival corticosteroids (Dexart®, Fuji Pharma Co., Japan) (1.65 mg/0.5 mL) given on the same day could not control the intraocular inflammation. Therefore, 25-G pars plana vitrectomy (PPV) was performed to rule out infectious endophthalmitis.

During 25-G PPV, we observed total tractional retinal detachment caused by extensively proliferative tissue between 3 and 9 o’clock. The intraocular lens was removed to improve the visibility of the fundus. When the proliferative tissue was peeled and removed with vitreous cutter and internal limiting membrane forceps, white masses appeared from the inside of the thick proliferative membrane (Fig. 2). These white masses were confirmed to be retained nuclear fragments and extracted from the superior corneoscleral flap. The fragments were approximately 60% in size of an ordinary lens nucleus (online suppl. Video; for all online suppl. material, see www.karger.com/doi/10.1159/000508913; the Supplemental Digital Content 1...
demonstrates the removal of lens fragments from the capsule formed by intravitreal proliferative tissue).

Bacterial culture test results were negative for both the aqueous humor and the nuclear fragments collected during the surgery. Based on a comprehensive assessment of these results and intraoperative findings, the patient was diagnosed with PVR secondary to LIU. Intraocular inflammation was reduced with postoperative oral and topical administration of steroid. Oral steroids were started at 30 mg/day, gradually decreased, and used for approximately 2 months. At 3 months after the initial surgery in our hospital, PPV and an encircling procedure were performed for the treatment of the remaining tractional retinal detachment. At 5 months after the second surgery in our hospital, transscleral suture of intraocular lens was performed. At 1 year following the initial visit to our hospital, the BCVA and intraocular pressure in the right eye was 0.1 and 12 mm Hg, respectively. In addition, intraocular inflammation was resolved (Fig. 3a), and the retina remained reattached (Fig. 3b).

**Discussion and Conclusion**

Recent sophistication of devices and techniques has made cataract surgery safer; however, complications have not been completely eliminated. Especially, surgery for hypermature cataract, like the present case, is difficult and therefore involves a high risk of complications. LIU, which occurs in up to 20% of cataract surgeries [2], is reported to be associated with a wide variety of ocular symptoms such as conjunctival injection, mutton-fat keratic precipitates, anterior chamber fibrin formation, hypopyon, retinal arteritis, macular edema, ocular hypertension, retinal detachment, and proliferative vitreoretinopathy [1, 2]. Therefore, if retained lens fragments are identified during the surgery, they must be processed thoroughly.

Although LIU develops after a complicated cataract surgery [3], it is crucial to differentiate LIU from postoperative infectious endophthalmitis and other forms of uveitis. Despite infectious endophthalmitis typically involving rapidly worsening intraocular inflammation, it is sometimes difficult to differentiate [6]. The present case did not demonstrate a sufficient response to steroid therapy, and postoperative infectious endophthalmitis could not be ruled out. Because it was necessary to confirm the status of the fundus, we chose to perform PPV, which enabled us to definitively diagnose LIU and perform suitable treatment. This result suggests that proactive PPV can be a treatment option when LIU is difficult to differentiate from postoperative infectious endophthalmitis.

In the present case, although the presence of retained lens fragments was not confirmed prior to the initial PPV, lens fragments were later removed from a capsule formed by proliferative tissue. Several reports have discussed how to manage dislocation of the lens nucleus into the vitreous cavity [7–13]. Only one of these reports indicated that “early” vitrectomy (i.e., less than 17 days after phacoemulsification) should be avoided [13], whereas many others advocate the superiority of early vitrectomy in removing the retained lens fragments [7–12]. Additionally, Chen et al. [14] have reported that severe complications occur more frequently when the interval between cataract surgery and vitrectomy is longer. In the above-cited study, PVR occurred in 55% of patients (n = 6/11) in whom the interval between cataract surgery and vitrectomy was longer than 1 week [14]. Our patient did not undergo PPV until 7 months after his initial cataract surgery and did not control postoperative intraocular inflammation during the follow-up period. This delay, as well as the persistent intraocular inflammation, may have led to pronounced PVR with tractional retinal detachment, and capsule formation around the retained lens fragments by proliferative membranes, thus making the patient’s condition...
refractory. These consequences highlight the need to consider early PPV to differentiate LIU from other diseases when uveitis of unknown origin is observed following cataract surgery with complications.

Interestingly, the patient’s blood test was negative for HLA-B51, but positive for HLA-A26. HLA-B51 and HLA-A26 are independent susceptibility genes involved in the onset of Behcet’s disease, and in particular, HLA-A26 is known as a risk factor for Behcet’s ocular lesions [15]. In this case, although there were no other systemic symptoms and Behcet’s disease could not be diagnosed, HLA-A26 was positive, which suggested a background of vascular inflammation. It is likely that abnormal, severe, and long-term intraocular inflammation occurred due to cataract surgery with complications for the eye with such a background. In the future, careful attention should be paid to intraocular surgery in patients who are positive for factors related to vascular inflammation, such as HLA-A26 and HLA-B51.

In conclusion, we report a case of LIU resulting in PVR. When uveitis develops following cataract surgery with complications, the possibility of LIU must be considered.

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Statement of Ethics

Consent to publish the case report was obtained. Moreover, this report does not contain any personal information that could lead to the identification of the patient in accordance with the Declaration of Helsinki.

Conflict of Interest Statement

There are no conflicts of interest.

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Author Contributions

Conception and design of the study: Hisanori Imai, Saki Inoue.
Acquisition of data: Akira Tetsumoto, Saki Inoue, Keiko Otsuka.
Analysis and/or interpretation of data: Hisanori Imai, Akira Tetsumoto, Saki Inoue.
Drafting the manuscript: Saki Inoue, Hisanori Imai.
Revising the manuscript critically for important intellectual content: Hisanori Imai, Makoto Nakamura.
All authors read and approved the final manuscript.
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Fig. 1. Findings at the first visit. a Slit-lamp photograph shows severe anterior chamber inflammation with hypopyon. b Optical coherence tomography shows tractional retinal detachment (yellow asterisk) and the extension of the proliferative membrane to the macula (white arrow).
**Fig. 2.** Intraoperative photography showing white masses (red arrow) covered with thick proliferative membrane (green arrow).

**Fig. 3.** Findings at the last visit. 
- Slit-lamp photograph shows improvement of intraocular inflammation.
- Optical coherence tomography shows that the retina was reattached.