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IDIOPATHIC PENETRATION OF CILIA INTO THE POSTERIOR SEGMENT PRESENTING AS SECTORAL SCLERITIS WITH PROGRESSIVE INTRAOCULAR INFLAMMATION

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Purpose: To report two cases of idiopathic intraocular cilia presenting as sectoral scleritis with progressive intraocular inflammation.

Methods: Both patients were treated with intravitreal antibiotics and underwent pars plana vitrectomy where the cilia were removed and identified on histopathology.

Results: One patient developed a retinal detachment while being treated for presumed endophthalmitis. The intraocular cilium was discovered during pars plana vitrectomy. In the second case, the cilium was detected on dilated fundus exam and was believed to be the cause of the patient's scleritis and vitritis. Therapeutic vitrectomy was performed. In both cases, the cilia were positively identified on histopathology.

Conclusion: Idiopathic intraocular penetration of cilia should be considered in the differential diagnosis of sectoral scleritis with progressive intraocular inflammation.

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Intraocular cilia have been widely reported in the setting of penetrating eye trauma and after ophthalmic surgery.¹ Less commonly, idiopathic intraocular cilia (IIC) have been reported in the absence of ocular trauma or surgery. To date, there have been

six reported cases of IIC, all of which have been associated with ocular inflammation.^{2–7} Three of the cases presented with scleritis and signs of endophthalmitis, such as vitritis or hypopyon.^{3,4,7} We hypothesize that IIC enter the eye by passage through the sclera, resulting in focal scleritis. Subsequent inoculation of the ocular fluids then results in intraocular inflammation.

We report two cases of IIC that follow a similar pattern of ocular inflammation. Our cases and a review of the literature suggest IIC should be included in the differential diagnosis of scleritis, particularly when refractory to anti-inflammatory treatments and associated with progressive intraocular inflammation.

Case Reports

Case 1

A 71-year-old man with no past medical or ocular history presented to the emergency department with left eye redness, pain,

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and vision loss. His symptoms had started 11 days before. He initially developed sectoral scleritis on the nasal aspect of the eye. Multiple interventions by the referring doctor were ineffective, including topical prednisolone acetate, oral ibuprofen, and oral prednisone 80 mg daily. He presented with worsening eye pain, headache, and “loss of all vision in the eye.”

The patient’s review of systems was positive for osteoarthritis of the wrists and infrequent oral ulcers. He was not using other medications and denied recreational drug use. He denied a history of ocular trauma or surgery.

Examination revealed visual acuity of 20/20 in the right eye and hand motions in the left eye. Intraocular pressure was normal in the right eye and 6 mmHg in the left eye. The left pupil was minimally reactive with no afferent pupillary defect. Slit-lamp examination of the left eye revealed nonblanching scleral injection nasally. No scleromalacia was noted in either eye. Anterior segment exam of the left eye was significant for a 1.3-mm hypopyon, 4+ cell, and 3+ flare. A membrane of fibrin covered the pupil. Significant media opacity precluded fundus examination.

B-scan ultrasonography demonstrated vitreous opacities consistent with vitritis. A focal elevation was noted in the nasal eye wall consistent with an abscess (Figure 1A). The retina was attached. The patient was treated empirically for endophthalmitis with vitreous tap and injection of vancomycin and ceftazidime. Fosfarnet was also administered intravitreally. The vitreous samples were sent for broad-range bacterial (16S ribosomal RNA gene sequencing) and fungal (28S ribosomal RNA gene and internal transcribed spacer sequencing) polymerase chain reaction, as well as viral polymerase chain reaction. Additional studies were obtained, including blood cultures, urine culture, HIV testing, rheumatoid factor, anti-PR3 antibody, anti-MPO antibody, anti-nuclear antibody, anti-CCP2 antibody, syphilis serology, QuantiFERON Gold, and chest x-ray. All tests were negative or noncontributory.

The patient was prescribed topical difluprednate, atropine, and ofloxacin. The oral prednisone was stopped. Five days after presentation, the work-up did not reveal an infectious etiology and he was started on oral prednisone 60 mg daily with subsequent taper. A ten-day course of oral moxifloxacin 400 mg daily was also prescribed. The inflammation in the left eye subsided. The view posteriorly remained limited because of vitreous haze.

One month after presentation, the patient developed a macula-involving rhegmatogenous retinal detachment. He underwent pars plana vitrectomy (PPV), scleral buckling, and silicone oil tamponade for repair. Intraoperatively, a subretinal cilium was discovered in the nasal retinal periphery, corresponding to the abscess visualized on B-scan ultrasonography (Figure 1B). The cilium was removed using intraocular forceps (Figure 1C, see also Video, Supplemental Digital Content 1, <http://links.lww.com/ICB/A103>, which demonstrates removal of intraocular cilium). The cilium was positively identified on histopathology. Repeat culture and polymerase chain reaction of the vitreous fluid showed no evidence of bacterial growth or bacterial DNA. The retinal detachment was believed to have originated from retinal necrosis adjacent to the cilium, which was barricaded with endolaser (Figure 1D).

Six months after his retinal detachment repair, he underwent repeat PPV for silicone oil removal, cataract extraction, and intraocular lens implantation. He subsequently developed an epiretinal membrane with chronic cystoid macular edema that responded partially to intravitreal triamcinolone. The patient’s best-corrected visual acuity 16 months after presentation was 20/60 and his retina remained attached (Figure 1E).

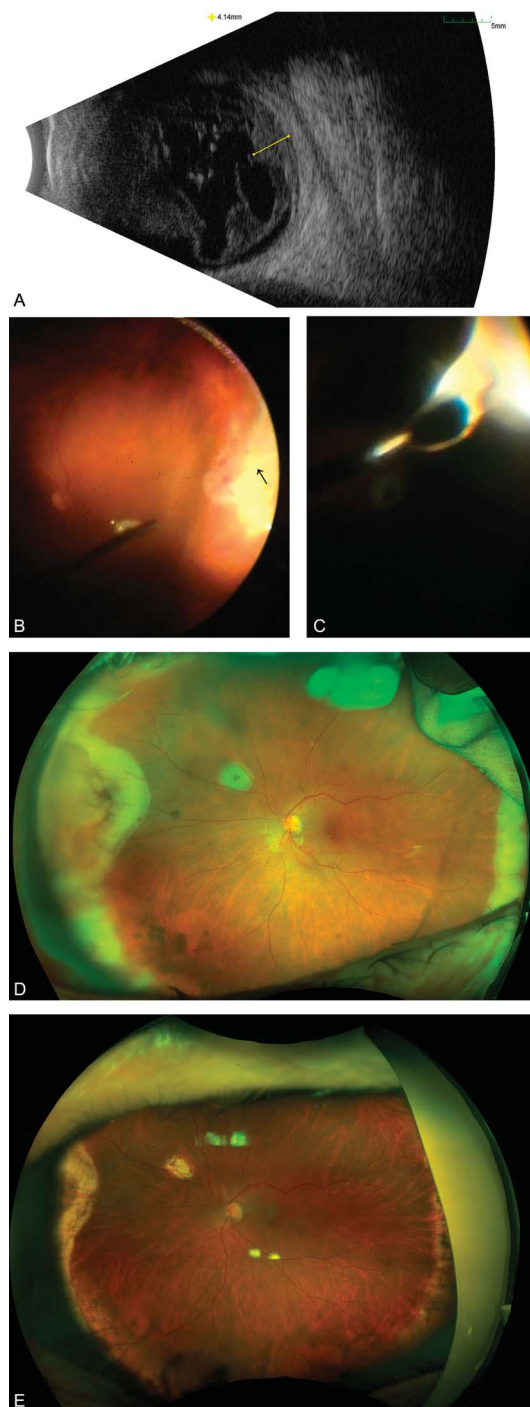


Fig. 1. A 71-year-old man with no past medical or ocular history presented with hypopyon uveitis and nasal sectoral scleritis. B-scan ultrasonography revealed an abscess underlying the sector of scleritis (A). The patient was treated with intravitreal antibiotics. One month later, he developed a rhegmatogenous retinal detachment that was repaired with PPV. Intraoperatively, a subretinal foreign body was observed in the nasal sector (B). The foreign body was removed with intraocular forceps (C) and found to be a cilium. The affected area was laser barricaded as seen in a postoperative week one widefield fundus photograph (D). The patient’s retina remained attached at postoperative month 8 as shown in the widefield fundus photograph, 2 months after silicone oil removal (E).

Case 2

A 43-year-old woman originally from Turkey was referred for evaluation of anterior scleritis of the right eye of 1-month duration. She had been managed by an outside provider with oral indomethacin and topical prednisolone acetate, which provided minimal relief. She had no past ocular history, denying previous trauma or surgery. Her medical history was significant for chronically low blood pressure. She was not using other medications. A complete review of systems was negative and the patient denied joint pain, skin rash, oral ulcers or genital ulcers.

Examination revealed a visual acuity of 20/15 in both eyes with normal intraocular pressures. Slit-lamp examination of the right eye revealed nonblanching scleral injection temporally with a small associated nodule (Figure 2A). No scleromalacia was noted in either eye. The right eye was otherwise quiet with no anterior chamber cell or flare. Dilated fundus exam in the right eye revealed an intraocular cilium underlying the focus of scleritis. One end of the cilium was embedded in the eye wall and the other end extended into the vitreous cavity. There was white inflammatory material forming a cap on the end of the cilium (Figure 2B). No cells were noted in the anterior vitreous. However, the cilium was visualized on B-scan ultrasonography with mild adjacent vitreous opacities (Figure 2C).

After 2 months observation, the patient developed 1+ anterior vitreous cell and complained of worsening floaters. There was enlargement of the cilium's inflammatory cap and increased adjacent vitreous opacities. The patient underwent PPV and removal of the cilium with intraocular forceps. Histopathology identified the structure as a hair shaft or cilium. The sector where the eyelash was retrieved was laser barricaded intraoperatively. The patient then received intravitreal vancomycin, ceftazidime, and dexamethasone. Vitreous cultures were negative. One month after vitrectomy, the patient's visual acuity in the right eye was 20/20 without further recurrence of scleritis or vitritis.

Discussion

Intraocular cilia in the setting of trauma or previous surgery have been described far more commonly than IIC. The clinical manifestations have been variable, including no signs of inflammation, vitreous bands forming from the cilium to the retina resulting in tractional retinal detachment, formation of iris cysts, corneal edema due to contact of the cilium with the corneal endothelium, or hypopyon

uveitis.^{1,8,9} In some cases, the cilium remained dormant in the eye for many years before causing any complications.¹

Idiopathic intraocular cilia have been reported six previous times in the literature, with all reports describing associated ocular inflammation (Table 1). The degree of inflammation has been variable, ranging from anterior uveitis, mild vitritis, up to culture-confirmed endophthalmitis. Of the six previously reported cases of IIC, three cases have reported sectoral scleritis at the onset of the course and another described "marked conjunctivitis."^{2-4,7} Both of the patients in our series had initially been managed at outside health care settings with presumed noninfectious scleritis. Idiopathic intraocular cilia should therefore be considered as a rare cause for refractory scleritis.

Whether the inflammatory process associated with IIC is secondary to inoculation of the eye with an infectious organism, mechanical irritation, or both is difficult to discern. Of the previously reported cases, four have resulted in vitreous cultures growing *Staphylococcal* species, suggesting an infectious contribution.^{3,4,7} Vitreous sampling in our cases did not yield confirmation of intraocular infection. However, the first case presented with hypopyon and severe vitritis that was clinically deemed to be endophthalmitis and responded favorably to intravitreal antibiotics.

All previous reports of IIC, including the two cases presented here, have required PPV and removal of the intraocular cilium.^{2-5,7} It is conceivable that a cilium may enter the eye without inciting intraocular inflammation. However, such a patient is unlikely to present for acute care. Excluding cases of previous ocular trauma or surgery, an intraocular cilium found asymptotically on routine exam has not to our knowledge been reported.

Rhegmatogenous or tractional retinal detachment has been a late complication of IIC in five of six previous cases.^{2-4,6,7} Four of the five cases ultimately resulted in the need for repeat PPV with or without scleral buckling.^{2-4,6} Three of the cases specifically

Fig. 2. A 43-year-old woman with no past ocular history or relevant medical history presented for management of anterior scleritis of the right eye as captured on slit-lamp photography (A). Fundus photography demonstrated a cilium partially buried in the eye wall and extending into the vitreous cavity with associated white inflammatory debris (B). B-scan ultrasonography redemonstrated the cilium buried in the eye wall (arrow heads) with adjacent vitreous opacities (arrow) (C).

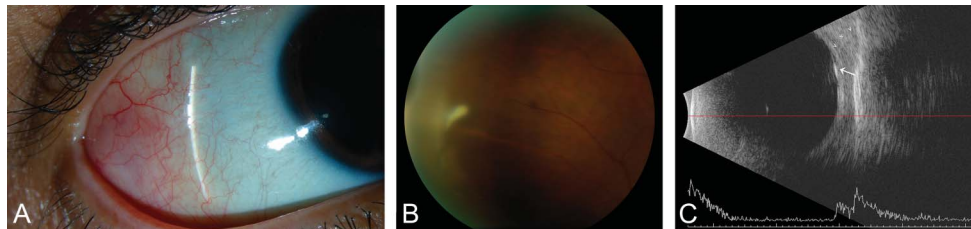


Table 1. Descriptions in the Literature of Idiopathic Intraocular Cilia

Publication	Report of Scleritis?	Description of Intraocular Inflammation	Detection of Cilium	Pars Plana Vitrectomy and Removal?	Culture Positive Endophthalmitis?	Course Complicated by Tractional or Rhegmatogenous Retinal Detachment?	Reported Visual Acuity at Presentation	Reported Visual Acuity at Final Follow-up
Oh et al ²	No, "marked conjunctivitis" described	Anterior segment cell, hypopyon	Observed on examination partially embedded in retina and extending into the vitreous, covered with whitish inflammatory debris; B-scan showed a linear hyper-echoic structure in the vitreous cavity with one end embedded in the retina	Yes	Yes, <i>Staphylococcus aureus</i>	Yes	20/125	Not reported
Kertes et al ³	Yes, temporal episcleral injection	Anterior chamber cell, anterior vitreous cell	Observed on examination partially embedded in retina and extending into the vitreous, covered with whitish inflammatory debris	Yes	No	Yes	20/30	20/20 at 18 months
Wirth and Helbig ⁴	Yes, acute scleritis in temporal sector	White vitreous inflammatory reaction	Observed a circumscribed white lesion in the posterior pole with white layering inflammatory material underneath, intraoperatively found to be a cilium; B-scan showed focal thickening of the sclera and choroid	Yes, with pathological evaluation	Yes, <i>Staphylococci</i>	Yes	20/20	20/15 at 1 year
Rossi et al ⁵	No	Trace anterior chamber cell, 1+ anterior vitreous cell	Observed "bright white wormlike object" free-floating in the anterior vitreous	Yes, with pathological evaluation	No growth from vitreous cultures	No	20/60	20/30 at 9 months

Table 1. (Continued)

Publication	Report of Scleritis?	Description of Intraocular Inflammation	Detection of Cilium	Pars Plana Vitrectomy and Removal?	Culture Positive Endophthalmitis?	Course Complicated by Tractional or Rhegmatogenous Retinal Detachment?	Reported Visual Acuity at Presentation	Reported Visual Acuity at Final Follow-up
Teo et al ⁶	No, "eye redness" described	Anterior uveitis, hypopyon, vitreous opacities	Not observed until vitrectomy, when removed	Yes, with pathological evaluation	Yes, <i>Staphylococcus aureus</i>	Yes	20/400	No light perception at 3 years
Jin et al ⁷	Yes, "temporal conjunctival and scleral injection"	Progressive worsening of anterior chamber cell, hypopyon, vitreous opacities, creamy intraretinal infiltrate	Not observed until vitrectomy, when removed	Yes, with pathological evaluation	Yes, <i>Staphylococcus aureus</i>	Yes	20/20	Not reported

mention the formation of membranes secondary to proliferative vitreoretinopathy.^{4,6,7} In our first case, the patient developed retinal detachment before the discovery of the intraocular cilium, which was treated similarly with PPV, scleral buckling, and silicone oil tamponade. No significant proliferative vitreoretinopathy was noted at the time of surgical repair, possibly because of prompt surgical intervention. Laser barricade was implemented in both of our cases to prevent future retinal detachment. Given the increased risk of proliferative vitreoretinopathy and redetachment in eyes with active inflammatory disease, it may be prudent to consider laser barricade, longer acting tamponade agents, or scleral buckling in eyes with IIC.

Although IIC is a rare entity, it should be considered in the differential diagnosis of sectoral scleritis with progressive intraocular inflammation. Evaluation of the vitreous and retina underlying the sector of scleritis may be revealing if there is an adequate view. In addition, B-scan ultrasonography can be used to visualize the cilium or associated abscess as described in our cases. Treatment with intravitreal antibiotics is likely beneficial if there are signs of endophthalmitis. Finally, efforts to prevent retinal detachment are warranted given a high number of reported cases requiring additional surgery to repair delayed-onset retinal detachment.

Key words: idiopathic intraocular cilia, cilium, eyelash, infectious scleritis, scleritis, endophthalmitis, intraocular foreign body, uveitis.

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