Case report

Bilateral recurrent macular holes

Stephen P. Yoon, Breanna A. Polascik, Dilraj S. Grewal, Sharon Fekrat

Abstract

Purpose: To report an unusual case of bilateral recurrent full-thickness macular holes (FTMH) in both eyes of a single patient over a 15-year period, with a total of 3 FTMH in the right eye and 2 in the left eye. Each FTMH was successfully treated with vitreous surgery, resulting in hole closure and visual acuity improvement.

Observations: During the previous 15 years, a 59-year-old female developed a total of 3 FTMH in the right eye and 2 FTMH in the left eye. The initial FTMH in each eye was surgically closed with pars plana vitrectomy (PPV), epiretinal membrane (ERM) peeling, 14% C3F8 gas placement, and face down positioning. Subsequent recurrences of FTMH, 2 in the right and 1 in the left, were surgically closed with PPV and ERM peeling and/or indocyanine green-assisted internal limiting membrane peeling, 14% C3F8 gas placement, and face down positioning. Seven years following the last FTMH surgical closure, the patient's best-corrected visual acuity was 20/50 in the right eye and 20/32 in the left eye with no FTMH in either eye.

Conclusions and importance: This case illustrates that a rare individual may have more than one recurrent FTMH in both eyes. Final visual outcome can be favorable following closure of more than one recurrent FTMH.

1. Introduction

Idiopathic full-thickness macular holes (FTMH) lead to loss of central vision and are most commonly caused by vitreous-mediated anteroposterior or tangential forces on the retinal surface. The prevalence of FTMH is estimated to be 3.3 per 1000, with 70% of patients being female. The majority develops a FTMH in only one eye and is able to maintain good visual acuity in the unaffected fellow eye. Chew and colleagues found that the rate of development of new macular holes in fellow unaffected eyes was low at 4.3% within 3 years of baseline examination. Reopening of a previously successfully operated macular hole is also uncommon with a reported rate of 4.8%–9.5%.

We report a unique case of bilateral recurrent full-thickness macular holes, 3 in the right eye and 2 in the left eye, each successfully treated with pars plana vitrectomy (PPV) resulting in hole closure and visual acuity improvement.

2. Case report

In 2002, a 59-year-old mildly myopic healthy female smoker presented with a two-month history of FTMH in the right eye. Best-corrected visual acuity (VA) was 20/80 in the right eye and 20/20 in the left eye. There was 2 + nuclear sclerosis in each eye. There was a posterior vitreous detachment (PVD) and a mild epiretinal membrane (ERM) in the right eye. She underwent a 20g pars plana vitrectomy (PPV) with ERM peeling and 14% C3F8 gas placement followed by two weeks of face down positioning. No indocyanine green (ICG) staining was used. One month postoperatively, the macular hole was closed and a subfoveal lucency was present (Fig. 1A). The VA improved to 20/40 in the right eye at 3 months with resolution of the subfoveal lucency (Fig. 1B). In 2003, a FTMH developed in the left eye with a decrease in VA to 20/50. A PVD and an ERM were present. She underwent a 20g PPV with ERM peeling and 14% C3F8 gas placement followed by two weeks of face down positioning. No ICG staining was used. One month postoperatively, the macular hole closed and a subfoveal lucency was present (Fig. 2A). The VA improved to 20/25 over time with resolution of the lucency in the left eye (Fig. 2B).

Due to progression of cataracts following the vitrectomy, she underwent uneventful cataract extraction and intraocular lens implantation in both eyes in 2004–5, and vision improved to 20/25 in both eyes.

In 2006, 3 years after successful FTMH closure, the left eye developed a recurrent FTMH associated with recurrent ERM formation (Fig. 2C). VA decreased to 20/64. She underwent 20g PPV with ERM peeling and 14% C3F8 gas placement followed by two weeks of face down positioning. No ICG staining was used. One month postoperatively, the macular hole closed and a subfoveal lucency was present (Fig. 2A). The VA improved to 20/25 over time with resolution of the lucency in the left eye (Fig. 2B).

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In 2007, 5 years after successful FTMH closure, a recurrent FTMH developed in the right eye (Fig. 1C), and VA decreased to 20/64. No ERM was present. She underwent 20g PPV with ICG-assisted peeling of the ILM, and 14% C3F8 gas placement followed by two weeks of face down positioning. One month postoperatively, the hole was closed (Fig. 1D), and the VA improved to 20/50. VA further improved to 20/32 in each eye without correction in 2008.

In 2010, 3 years after successful FTMH closure, she developed a recurrent FTMH with a recurrent ERM in the right eye (Fig. 1E), and VA dropped to 20/50. An intravitreal injection of triamcinolone followed by 0.3 cc of 100% C3F8 was administered in the office. The hole remained open. She underwent a 23g PPV in the right eye with ERM peeling. ICG staining revealed no ILM. 14% C3F8 gas was placed followed by 10 days of face down positioning. One month postoperatively, the hole was closed. VA improved to 20/50. VA further improved to 20/32 in each eye without correction in 2008.

At her most recent visit in 2017, fifteen years following the first macular hole, VA was 20/50 in the right eye and 20/32 in the left eye with no further recurrences of the macular hole in either eye (Figs. 1G and 2E).

3. Discussion

The formation of bilateral idiopathic FTMH is a relatively rare clinical phenomenon. Ezra et al. conducted the first prospective study of the incidence of idiopathic FTMH in the fellow eye in 1998 and reported a 7.5% involvement rate at 18 months and 15.6% involvement rate at five years in previously unaffected fellow eyes without PVD.2 Chew et al. performed a prospective study without accounting for presence of PVD at baseline and reported lower rates of involvement of the fellow eye: 4.3% within 3 years, 6.5% at 4–5 years, and 7.1% at 6 or more years.3 The mean interval time between onset of the first FTMH to the onset of a macular hole in the fellow eye was found to be 26.1 months with no significant difference between men and women in a retrospective study by Kumagai et al.6 The presence of predisposing foveal lesions, such as an impending hole in the fellow eye, increases the risk of progression to FTMH by 40–60%.1 The risk of progression to FTMH in the fellow eye with a preexisting PVD has been reported to be < 1%.7

After initial successful surgical repair of FTMH in both eyes, our patient presented with two recurrences in the right eye and one in the left eye over a 15-year time period. Reopening is an uncommon outcome of a successfully closed FTMH, and the exact mechanism of reopening is not well understood. ILM peeling is now widely used in macular hole surgery, with reduction in the rate of reopening from 2% to 16% in eyes in which the ILM was not peeled to 0–8.6% in eyes in which the ILM was peeled.1 The lower rate of reopening after ILM peeling suggests that ILM contracture may be involved in the reopening process. Our patient did not undergo ILM peeling with the first PPV in
each eye in the early 2000’s, which may have increased her risk of recurrence after initial surgical closure. Our patient had an associated ERM in four of the five macular holes. A less common pathway of FTMH formation is through tangential ERM traction, usually on a lamellar hole.8 Our patient had a stage IV FTMH with an ERM upon initial presentation in each eye, which makes it difficult to determine if the FTMH occurred before the onset of ERM or as a result of it. The absence of a retinal configuration suggesting a preexisting inner tissue defect on OCT and the lack of lamellar hole-associated epiretinal proliferation make an ERM-mediated mechanism less likely.9 However, the presence of an ERM during FTMH recurrence in our patient may indicate that Müller cells and fibrous astrocytes involved in sealing the retinal defect after PPV continued to proliferate and contracted. The tangential traction on the foveal area exerted by these cells may have contributed to the multiple recurrences of FTMH.

Our patient underwent cataract extraction in both eyes after initial successful surgical closure in both eyes. The role of cataract extraction in the reopening of macular holes remains a topic of controversy. Bhatnagar et al.10 observed that pseudophakic cystoid macular edema (CME), a possible consequence of cataract extraction, was associated with a seven-fold increased risk of macular hole reopening. Paques et al.5 reported a possible association between reopening and uneventful cataract extraction in patients without the development of pseudophakic CME. They hypothesized that cataract extraction may lead to the passage of chemotactic factors to glial cells and the formation of an ERM over the operated macular hole. The association between cataract extraction and the recurrence of FTMH remains inconclusive due to the high rate of cataract extraction following surgical closure of FTMH and the overall low number of reported cases with reopening.8

Reoperation after reopening has favorable anatomic and visual outcomes. Christmas et al.4 reoperated on 12 eyes and described the mean visual acuity after reoperation being comparable to visual acuity before reopening. In their study, final visual outcome was not dependent on the interval between surgical closure and reopening. Hussin et al.11 described a case of three recurrences of a single macular hole after initial successful surgical closure with a favorable visual outcome following multiple repeat macular hole operations. This case was not associated with any of the potential risk factors implicated in the reopening of macular holes.

In conclusion, we report a rare case of a patient with bilateral recurrent FTMH who underwent five total PPVs between both eyes, 3 in the right eye and 2 in the left eye over a follow up period of 15 years and has maintained good visual outcomes after each surgical closure. While further studies are necessary to elucidate the factors that may be involved in the recurrence of FTMH, our case demonstrates that final visual outcome can be favorable following multiple surgeries for bilateral recurrent FTMH.

4. Patient consent

The patient consented to publication of the case orally. This report does not contain any personal information that could lead to the identification of the patient.

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Conflicts of interest

The authors do not have any propriety interest or conflicts of interest.

Authorship

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

1. Ezra E. Idiopathic full thickness macular hole: natural history and pathogenesis. Br J Ophthalmol. 2001;85:102–108.
2. Ezra E, Wells JA, Gray RH, et al. Incidence of idiopathic full-thickness macular holes in fellow eyes. A 5-year prospective natural history study. Ophthalmology. 1998;105:353–359.
3. Chew EY, Sperduto RD, Hiller R, et al. Clinical course of macular holes: the eye disease case-control study. Arch Ophthalmol. 1999;117:242–246.
4. Christmas NJ, Smiddy WE, Flynn Jr HW. Reopening of macular holes after initially successful repair. Ophthalmology. 1998;105:1835–1838.
5. Paques M, Massin P, Blain P, Duquesnoy AS, Gaudric A. Long-term incidence of reopening of macular holes. Ophthalmology. 2000;107:760–765 discussion 6.
6. Kumagai K, Ogino N, Hangai M, Larson E. Percentage of fellow eyes that develop full-thickness macular hole in patients with unilateral macular hole. Arch Ophthalmol. 2012;130:393–394.
7. Kumagai K, Furukawa M, Ogino N, Larson E. Incidence and factors related to macular hole reopening. Am J Ophthalmol. 2010;149:127–132.
8. Tsai CY, Hsieh YT, Yang CM. EPIRETINAL membrane-induced full-thickness macular holes: the clinical features and surgical outcomes. Retina. 2016;36:1679–1687.
9. Fekrat S, Wendel RT, de la Cruz Z, Green WR. Clinicopathologic correlation of an epiretinal membrane associated with a recurrent macular hole. Retina. 1995;15:53–57.
10. Bhatnagar P, Kaiser PK, Smith SD, Meisler DM, Lewis H, Sears JE. Reopening of previously closed macular holes after cataract extraction. Am J Ophthalmol. 2007;144:252–259.
11. Hussin HM, Asaria RH, Knox Cartwright NE, Grey RH. Fourth time lucky: a case of multiple recurrence of a macular hole. Br J Ophthalmol. 2006;90:659.