Contents lists available at ScienceDirect



American Journal of Ophthalmology Case Reports

journal homepage: www.elsevier.com/locate/ajoc

Case report

Spontaneous closure of a chronic full thickness macular hole after failed surgery



AMERICAN URNAL OI OPHTHALMOLOGY

CASE REPORTS 壳

Jennifer Lee, Vincent Q. Nguyen, Mallika K. Doss, Andrew W. Eller*

Retina Service, Department of Ophthalmology, University of Pittsburgh Medical Center, Pittsburgh, PA, USA

ARTICLE INFO ABSTRACT Keywords: Purpose: To describe an unusual case of spontaneous closure of a chronic, large, idiopathic, stage 4 macular hole

Idiopathic macular hole Spontaneous closure Optical coherence tomography Polypoidal choroidal vasculopathy Type 1 neovascularization

after failed surgery.

Observations: A 75-year-old female presented with a history of a chronic, full thickness macular hole after failed surgery in the right eye. Two years after onset, she developed a fibrotic scar, which closed the macular hole and unexpectedly improved her vision. At her 4 year follow up exam, optical coherence tomography demonstrated a stable, closed macular hole with continued improvement in her visual acuity despite lack of surgical and medical intervention.

Conclusions and importance: The spontaneous closure of an idiopathic full thickness macular hole is an unusual event. When it occurs, it is typically in an acute setting and is attributed to bridging retinal tissue, vitreofoveal separation, and a small diameter size. In this report, we show that a chronic, large break, that failed prior surgical intervention, can spontaneously close. The formation of an underlying fibrotic scar from type 1 neovascularization bridged the macular hole and improved her visual acuity.

1. Introduction

Macular holes are retinal breaks in the fovea involving the partial to complete dehiscence of the neural retinal layers.¹ Formation of an idiopathic or primary macular hole has been attributed to mechanical forces exerting tangential traction at the vitreomacular interface. The most advanced state of a macular hole is stage 4, described as a full thickness macular hole (FTMH) with complete posterior vitreous separation.

Spontaneous closure is a rare event.^{2–4} When it occurs, the macular hole usually has an inciting cause such as trauma,⁵ laser procedure,⁶ or intraocular surgeries.^{7–9} We present a case of spontaneous closure of an idiopathic stage 4 FTMH with the following unique features: large size, chronic history, history of failed surgical intervention, and association with type 1 neovascular tissue.

1.1. Case report

A 75-year-old African-American female with a history of a chronic FTMH in her right eye presented for a retinal evaluation. Her surgical history was significant for right eye pars plana vitrectomy with internal limiting membrane (ILM) peeling and C3F8 gas tamponade that was performed one year earlier. On her initial presentation to our clinic, she reported new floaters in her left eye for the past two months.

Her best corrected visual acuity was 20/400 in the right eye and 20/ 40 in the left eye. Slit-lamp exam was notable for posterior chamber intraocular lens in both eyes. Examination of the right fundus showed a FTMH, epiretinal membrane, with an arc of retinal degeneration at the level of the retinal pigment epithelium (RPE) along the temporal border of the optic nerve. Her fluorescein angiography demonstrated late phase RPE staining corresponding to the area of the macular hole and peripapillary retinal degeneration (Fig. 1). In the left fundus, there were intraretinal hemorrhages along the superior arcade extending to the fovea. Optical coherence tomography (OCT) of the right eye confirmed a stage 4 FTMH (Fig. 2A). The aperture measured 525 µm, which categorized this hole as large in size (International Vitreomacular Traction Study¹). OCT of the left eye revealed cystoid macular edema secondary to a branch retinal vein occlusion. Management of her left eye included sectoral scatter photocoagulation followed by a series of 3 intravitreal bevacizumab injections. She declined additional surgery for the right eye.

Nine months after her initial visit, OCT of the right eye revealed a new fibrotic scar secondary to spontaneous regression of type 1 neovascularization. The macular hole was found to be closed (Fig. 2 B) and her best corrected visual acuity improved to 20/200. Similarly, OCT of her left eye revealed type 1 neovascularization with pigment epithelial

https://doi.org/10.1016/j.ajoc.2018.12.006

Received 7 August 2018; Received in revised form 2 December 2018; Accepted 4 December 2018 Available online 05 December 2018

2451-9936/ © 2018 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/BY-NC-ND/4.0/).

^{*} Corresponding author. The Eye and Ear Institute, 203 Lothrop Street, Pittsburgh, PA, 15213, USA. E-mail address: elleraw@upmc.edu (A.W. Eller).



Fig. 1. Color fundus photo of the right eye (top image) demonstrating a full thickness macular hole, epiretinal membrane, and an arc of retinal degeneration at the level of the retinal pigment epithelium along the temporal border of the optic nerve. Corresponding late phase fluorescein angiography demonstrating RPE staining (bottom image). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

detachment and overlying intraretinal fluid, which was treated with intravitreal bevacizumab. Two years after her initial visit, her right eye visual acuity continued to improve to 20/70 and OCT re-demonstrated a closed macular hole with decreased cystoid edema (Fig. 2C). At her three and four year follow-up, her OCT showed resolution of intraretinal cystoid edema, stable disciform scar, with a visual acuity of 20/50-2 at both visits (Fig. 2 D and E). Serial en face images at the level of the inner segment/outer segment-ellipsoid junction revealed progression of hyperreflective material from the peripapillary region towards the fovea (Fig. 2 A-E, top images). Throughout this time, she had no treatment of her right eye and a total of seven injections of bevacizumab in the left eye.

2. Discussion

Spontaneous closure of stage 4 idiopathic full thickness macular holes are unusual events. When it occurs, it is typically noted within 1–6 months of onset.^{10–12} Previously, the pathophysiology behind this process has been attributed to the formation of bridging intraretinal tissue.¹² Closure is also more likely when the breaks are small, with an aperture size of less than 250 μ m.^{1,10,11} Moreover, a literature search failed to identify cases of spontaneous closure following pars plana vitrectomy with ILM peeling. In our case, we not only describe a chronic large hole, but one that closed two years after failed surgical intervention.

While the development of neovascular tissue serving as a nidus for macular hole closure has been previously reported, it was associated with age-related macular degeneration and the patient had significant visual acuity improvement after intravitreal bevacizumab.¹³ In distinction to this report, our patient presumably had polypoidal choroidal

vasculopathy (PCV) given her race, presence of peripapillary atrophy, and lack of drusen on exam. Moreover, she had progressive visual acuity improvement without treatment. Although this is highly unusual, the presumed PCV appears to exist entirely below the RPE and given her vision, there must be preservation of the outer retinal layers. This speculation is supported by the thickness of the outer nuclear layer, which contains nuclei of photoreceptors. Qualitatively, the preserved thickness of the layer over time serves as an indicator of photoreceptor integrity and can explain her visual acuity outcome.

Macular holes develop as a result of either tangential traction on the surface of the retina or from anteroposterior traction from the vitreous. These mechanisms are addressed during surgical repair with release of vitreous traction when present, and peeling of the ILM. More recently, larger macular holes have been successful closed with placement of an ILM flap over the hole.^{14,15} There is extensive literature on various methods to treat myopia-associated retinal detachments secondary to macular holes using a macular scleral buckle.¹⁶ The role of the buckle is to counteract traction on the retina. In this case report, the subretinal disciform scar may have served as "nature's" scleral buckle beneath the macular hole. This suggests that one may consider the placement of a macular scleral buckle for treatment of recalcitrant cases.

3. Conclusion

In summary, we report a rare case of spontaneous closure of a chronic, stage 4, macular hole due to type 1 neovascularization. We have followed this event clinically and tomographically and confirmed the stability of her macular hole closure over time. In addition, our patient is unusual because she does not share the same characteristics as other patients who had similar outcomes in terms of her visual acuity improvement, size of break, chronicity, and prior surgical intervention.

Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

Funding

This work was supported by NIH CORE Grant P30 EY08098 to the Department of Ophthalmology, the Eye and Ear Foundation of Pittsburgh, and from an unrestricted grant from Research to Prevent Blindness, New York, NY.

Conflicts of interest

The following authors have no financial disclosures: JL, MD, VN, AE.

Authorship

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

Acknowledgements

None.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ajoc.2018.12.006.



Fig. 2. A-E. Serial horizontal (bottom images) and en face (top images) optical coherence tomography (OCT) images at the level of the inner segment/outer segment-ellipsoid junction. **A** At the initial examination, a full-thickness hole with perifoveal cystoid degeneration and a visual acuity of 20/400. **B** Nine months later, the hole is closed through the formation of an underlying fibrotic scar, her vision improved to 20/200. **C** Two years later, she had decreased cystoid edema and

her visual acuity improved to 20/70. **D** Three years later, she had complete resolution of intraretinal fluid and continued visual acuity improvement to 20/50-2. **E** Four years later, re-demonstration of a closed macular hole with stable disciform scar and a vision of 20/50-2.

References

- Ophthalmology. 2013;120(12):2611–2619.
 Ferris JD. Pathogenesis of idiopathic macular holes. *Curr Opin Ophthalmol.* 1997;8(3):87–93.
- 3. Gass JD. Idiopathic senile macular hole. Its early stages and pathogenesis. Arch Ophthalmol. 1988;106(5):629–639 Chicago, Ill. : 1960.
- 4. la Cour M, Friis J. Macular holes: classification, epidemiology, natural history and treatment. Acta Ophthalmol Scand. 2002;80(6):579–587.
- Yamashita T, Uemara A, Uchino E, Doi N, Ohba N. Spontaneous closure of traumatic macular hole. *Am J Ophthalmol*, 2002;133(2):230–235.
- Park J, Son Y, Lee S. Macular hole formation and spontaneous closure following neodymium-doped yttrium aluminum garnet capsulotomy in a vitrectomized eye. Indian J Ophthalmol. 2016;64(2):165–166.
- Bhojwani D, Vasavada S, Sudhalkar A, Vasavada V, Vasavada AR. Acute full-thickness macular hole after uneventful femtosecond-assisted cataract surgery and its spontaneous closure. *Indian J Ophthalmol.* 2018;66(6):848–849.
- Cohen MN, Baumal CR. Macular hole formation followed by spontaneous closure after pneumatic retinopexy in a patient with diabetic macular edema. *Retin Cases Brief Rep.* 2018:11 [Epub ahead of print].
- 9. Tadayoni R, Massin P, Haouchine B, Cohen D, Erginay A, Gaudric A. Spontaneous

resolution of small stage 3 and 4 full-thickness macular holes viewed by optical coherence tomography. *Retina*. 2001;21(2):186–189.

- Hamano R, Shimoda Y, Kishi S. Tomographic features of spontaneous closure of fullthickness macular holes. Jpn J Ophthalmol. 2007;51(1):76–77.
- Gonzalez-Cortes JH, Toledo-Negrete JJ, Bages-Rousselon Y, de Alba-Castilla MA, Mohamed-Hamsho J. Spontaneous closure of simultaneous idiopathic macular holes documented by spectral-domain optical coherence tomography. *Retin Cases Brief Rep.* 2018.
- Imasawa M, Chiba T, Iijima H. Spontaneous closure of idiopathic full-thickness macular holes in both eyes. Jpn J Ophthalmol. 2010;54(5):507–509.
- Su D, Klufas MA, Hubschman JP. Spontaneous closure of a full-thickness macular hole with conversion to exudative age-related macular degeneration. JAMA Ophthalmol. 2016;134(5):604–606.
- Leisser C, Hirnschall N, Doller B, et al. Internal limiting membrane flap transposition for surgical repair of macular holes in primary surgery and in persistent macular holes. *Eur J Ophthalmol.* 2018;28(2):225–228.
- Michalewska Z, Michalewski J, Adelman RA, Nawrocki J. Inverted internal limiting membrane flap technique for large macular holes. *Ophthalmology*. 2010;117(10):2018–2025.
- 16. Alkabes M, Mateo C. Macular buckle technique in myopic traction maculopathy: a 16-year review of the literature and a comparison with vitreous surgery. Graefe's archive for clinical and experimental ophthalmology = Albrecht von Graefes Archiv fur klinische und experimentelle Ophthalmologie. 2018;256(5):863–877.