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Spontaneous closure of an idiopathic macular hole after epiretinal membrane development

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ABSTRACT

Purpose: To report non-operative closure of an idiopathic full thickness macular hole (FTMH) spontaneous secondary to the development of a macular epiretinal membrane (ERM).

Observations: A 68-year-old woman, with no relevant medical history, and a 6-month history of decreased visual acuity in her right eye was diagnosed to have an idiopathic FTMH. The patient refused surgery and the FTMH was followed-up for seven years. The spectral domain optical coherent tomography follow-up showed the evolution of the FTMH and its spontaneous closure after development of an ERM. In the presence of an ERM with vitreo-papillary detachment, it is possible that the centripetal forces involved helped bring together the edges of the macular hole resulting in a possible spontaneous closure. Additionally and separately, the presence of an ERM may act as scaffolding for Muller cell migration and consequent macular hole closure.

Conclusions and importance: Development of an ERM was followed by non-operative FTMH closure in this specific case. It is important to note, that this is an extraordinary situation in which the patient had a favorable anatomical evolution despite having rejected conventional surgical intervention. Studies aimed at determining the mechanisms and situations in which these cases occur could provide answers that help us make more appropriate decisions. To our knowledge, the present case is the first in the literature to report a spontaneous closure of a FTMH secondary to the appearance and progression of a previously non-existent ERM.

1. Introduction

A full thickness macular hole (FTMH) is a defect located in the central fovea that can partially or totally compromise the neurosensorial retina.[1] The incidence and prevalence of MH varies considerably. McCannel et al.,reported the incidence to be 7.8 per 100,000 person--years.[2] The reported prevalence is 0.2/1000 in Australia and 3.3/1000 in the United States.[3,4] FTMHs are usually unilateral and tend to affect women more frequently between the sixth and seventh decade of life.[2] A diagnosis of FTMH is based on clinical examination and confirmed by optical coherence tomography (OCT).[5] Surgical closure is the standard of care and it is uncommon for FTMHs to close spontaneously. In the rare event of a spontaneous closure, its resolution is related to the size of the macular hole.[6–8] Even with several advances in imaging, the understanding of the precise mechanisms of MH closure remains limited.

An epiretinal membrane (ERM) is the consequence of cellular proliferation and accumulation of fibrous-like tissue over the internal surface of the retina.[9] ERMs are mainly idiopathic, but have been associated with pre-existing conditions or they can be iatrogenic in origin.[9,10] Multiple investigations describe the coexistence and pathophysiological relationship between FTMH and ERM. Posterior hyaloid tangential traction release and fibrous tissue proliferation may play a major role in spontaneous closure.[5,10,11]

The purpose of this article is to report the spontaneous closure of an idiopathic grade 4 FTMH, with minimal diameter greater than 400μm, secondary to the appearance and progression of a previously non-existent ERM documented by spectral domain OCT (SD-OCT).

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2. Case history

A 68-year-old female patient presented with a 6-month history of decreased vision, central scotoma and metamorphopsia in her right eye (RE), that was gradual, painless and progressive. There was no history of trauma and no other relevant ophthalmological or medical history.

The ocular examination demonstrated a best-corrected visual acuity (BCVA) of 20/80 and 20/30 for the RE and the left eye (LE) respectively. The anterior segment assessment was unremarkable except for an early senile cataract (NO2, NC2, C1, P1) in both eyes (OU).[12] Posterior segment evaluation revealed normal optic discs in both eyes and a Stage 4 macular hole in the RE.[13]

The macular hole dimensions were confirmed by SD-OCT (CIRRUS™ HD-OCT 5000). According to the International Vitreomacular Traction Study Classification System,[5] the MH was defined as a Grade 2B FTMH of 282μm (Fig. 1A). Surgical intervention was recommended, however the patient decided not to undergo the procedure for personal reasons. She however agreed for regular follow-up. Her follow-up visits were documented with SD-OCT images over 7.5 years (90 months and 18 days), as is shown in Fig. 1.

At the second year of follow-up, the patient still reported a central scotoma and metamorphopsia, her BCVA was of 20/400, the early senile cataract remained stable, and the SD-OCT showed progression of the MH to a Grade 4 MH (407μm) as is evident in Fig. 1C. For the next three years of follow-up, a slow progression of senile cataract was documented and SD-OCT findings remained stable (Fig. 1D).

In the seventh year of follow-up, we documented an ERM for the first time (Fig. 1E). Four months later, the patient stated improvement in her perception of the central scotoma and metamorphopsia. Her BCVA was 20/400, ocular examination revealed progression of the senile cataract (NO4, NC4, C1, P1) and of the ERM. The SD-OCT revealed progression of the ERM and spontaneous closure of the FTMH (Fig. 1F).

3. Discussion

The spontaneous closure of a FTMH is uncommon and the mechanisms through which it can occur are complex.[7] Most of the cases described in the literature of spontaneous closure refer mainly to macular holes of traumatic origin.[14] Zhang et al. described spontaneous closure of FTMH in 3 patients with MH developed after posterior vitrectomy and concluded that spontaneous closure is possible in small MHs in vitrectomized eyes despite the presence of ERM.[15] Smiddy et al. described a case of spontaneous closure of a FTMH after 2 years of follow-up, the closure of the FTMH was related to the progression of a preexisting ERM.[16] To our knowledge, the present case is the first in the literature to report a spontaneous closure of a FTMH secondary to the appearance and progression of a previously non-existent ERM in a grade 4 MH with minimal diameter greater than 400μm.

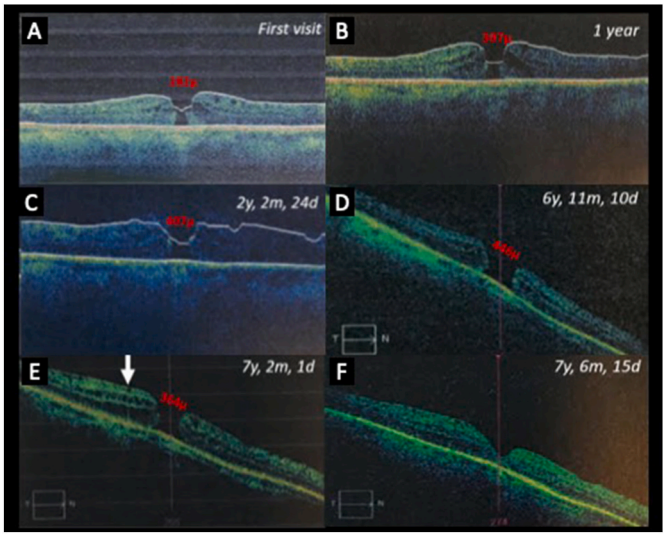

Fig. 1. SD-OCT images of the FTMH and ERM. **A**, shows a FTMH and no sign of an ERM. **B-D**, show progressive enlargement of the macular hole with a slight augmentation of the cystic spaces and lifting of its edges **(B–D)**. After seven years, the appearance of an ERM (white arrow) is shown in **(E)**. **F** show the presence of the ERM and the complete closure of the FTMH.

In the present case, we describe this occurrence as a spontaneous closure. The pathophysiological phenomena leading probably to spontaneous closure have been previously described by Sebag et al.[8] They state that when the posterior cortex of the vitreous divides, at a level posterior to the hyalocytes, a thin hypocellular membrane remains attached to the macula. If the posterior vitreous (PV) still attached to the optic disc, it may induce centrifugal tangential contraction and may cause a FTMH. When this separation leaves a layer of hyalocytes in the anterior macular area, an ERM may develop. Contraction of this tissue causes centripetal tangential traction upon the underlying retina and results in macular pucker, which generally occurs when the PV adhesion from the optic disc is released, which suggests the final stage of posterior vitreous separation.[8] On the other hand, there is evidence of remnant posterior cortex tissue noted during MH surgery, were a negative staining after brilliant blue dye injection indicated presence of tissue over the internal limiting membrane (ILM).[17]

The present case shows the coexistence of a FTMH and an ERM; however they did not occur simultaneously and this allowed us to observe the chronological behavior of this anomalous PV detachment. We propose the following hypothesis: Initially, the formation of the FTMH occurred as a consequence of the centrifugal forces associated with the vitreo-papillary adhesion (VPA) and its persistence. Later on, after the appearance of the ERM and with detachment of the VPA, centripetal forces were developed, narrowing the edges of the MH resulting in spontaneous closure. This is based on Sebag et al. observations were they stated that VPA may have an important influence on the vectors of force at the vitreoretinal interface, and that VPA is commonly seen in FTMHs compared to macular pucker and vitreo-papillar detachment is more frequently seen in macular pucker compared to FTMHs.[8,18] A second mechanism of closure (not unlike the one we describe but one that needs surgical intervention), has been described in reports of ILM grafts used for FTMH closure. Grafts may act as possible scaffolding between the edges of the hole favoring the migration of Muller cells and the consequent hole closure,[19] an occurrence that happened spontaneously in our case the ERM itself. Both pathophysiological mechanisms are possible explanations for the spontaneous closure of the FTMH in this particular case.

Petropoulos IK et al. described a case of FTMH spontaneous closure. They identified an ERM since presentation and according to the OCT scans the FTMH size looks small.[20] Our case is different in certain things. First, the spontaneous closure occurred in a large FTMH with minimal diameter of 407 μm. Secondly, the ERM developed after seven years of follow-up. Similarly, Michalewska et al., described two cases of stage III and stage IV MH spontaneous closure.[21] They described spontaneous closure of FTMH with a minimal diameter less than 300 μm and in absence of ERM which defer from our case.

It is important to note, that in the present case vision of 20/400 remained the same despite FTMH spontaneous closure. The closure occurred after seven years, in this period of time the early senile cataract (NO2 NC2 C1 P1) evolved to a (NO4 NC4 C1 P1) senile cataract. After the FTMH closure, foveal disorganization of the outer retinal layers is evident as in OCT scans suggestive of photoreceptor damage. Senile cataract and photoreceptor damage, both may explain the persistent 20/400 vision after FTMH spontaneous closure, however patient stated improvement in the central scotoma and metamorphopsia after FTMH closure. It is important to note that this is an extraordinary situation in which our patient had a favorable anatomical evolution despite having rejected conventional surgical intervention. The fact that in this

particular case our patient had a favorable anatomical evolution does not mean a change for surgical indications. Stage 2 or higher macular holes are good indications for surgical intervention.[22]

It is interesting to see the correlation of the FTMH and the ERM and how this could contribute to the closure of the hole. Studies aimed at determining the mechanisms and situations in which these cases occur could provide answers that help us to better understand vitreomacular pathologies.

Summary statement

The idea of our article is to present a case of spontaneous closure of a full thickness macular hole, with a minimal diameter greater than 400 μm, secondary to the development of an epiretinal membrane during a follow-up of seven years. We explore possible mechanisms for this event as well as the pathogenesis and vitreomacular dynamics.

Consent statement

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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Authorship

All the authors state that they meet the current ICMJE criteria for Authorship. Authors JG, AB, JP, AT, JGC conceptualized the manuscript and were involved in the collection, analysis and interpretation of data and in the final drafting and review of the manuscript. Author AS was involved in the analysis and interpretation of data and in the final drafting and review of the manuscript.

Ethical considerations

The manuscript data adheres to the tenets of Helsinki.

Declaration of competing interest

The authors declare no conflicts of interest in any concept or product described in this article. The authors declare no proprietary interest.

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References

1. Gass JDM. Idiopathic senile macular hole: its early stages and pathogenesis. *Arch Ophthalmol*. 1988;106(5):629–639.
2. McCannel CA, Ensminger JL, Diehl NN, Hodge DN. Population-based incidence of macular holes. *Ophthalmology*. 2009;116(7):1366–1369.
3. Mitchell P, Smith W, Chey T, Wang JJ, Chang A. Prevalence and associations of epiretinal membranes: the blue mountains eye study, Australia. *Ophthalmology*. 1997;104(6):1033–1040.
4. Rahmani B, Tielsch JM, Katz J, et al. The cause-specific prevalence of visual impairment in an urban population: the Baltimore Eye Survey. *Ophthalmology*. 1996; 103(11):1721–1726.
5. Duker JS, Kaiser PK, Binder S, et al. The International Vitreomacular Traction Study Group classification of vitreomacular adhesion, traction, and macular hole. *Ophthalmology*. 2013;120(12):2611–2619.
6. Meuer SM, Myers CE, Klein BEK, et al. The epidemiology of vitreoretinal interface abnormalities as detected by spectral-domain optical coherence tomography: the beaver dam eye study. *Ophthalmology*. 2015;122(4):787–795.
7. Yuzawa M, Watanabe A, Takahashi Y, Matsui M. Observation of idiopathic full-thickness macular holes: follow-up observation. *Arch Ophthalmol*. 1994;112(8): 1051–1056.
8. Sebag J, Gupta P, Rosen RR, Garcia P, Sadun AA. Macular holes and macular pucker: the role of vitreoschisis as imaged by optical coherence tomography/scanning laser ophthalmoscopy. *Trans Am Ophthalmol Soc*. 2007;105:121.
9. Wiese GN. Clinical features of idiopathic preretinal macular fibrosis. *Am J Ophthalmol*. 1975;79:349–357.
10. Appiah AP, Hirose T. Secondary causes of premacular fibrosis. *Ophthalmology*. 1989; 96(3):389–392.
11. 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*. 2021;15(1):27–30.
12. Chylack LT, Wolfe JK, Singer DM, et al. The lens opacities classification system III. *Arch Ophthalmol*. 1993;111(6):831–836.
13. Gass JDM. Reappraisal of biomicroscopic classification of stages of development of a macular hole. *Am J Ophthalmol*. 1995;119(6):752–759.
14. Yeshurun I, Guerrero-Naranjo JL, Quiroz-Mercado H. Spontaneous closure of a large traumatic macular hole in a young patient. *Am J Ophthalmol*. 2002;134(4):602–603.
15. Zhang W, Grewal DS, Jaffe GJ, Mahmoud TH, Fekrat S. Spontaneous closure of full-thickness macular hole with epiretinal membrane in vitrectomized eyes: case series and review of literature. *Ophthalmic Surgery, Lasers Imaging Retin*. 2017;48(2): 183–190.
16. Smiddy WE. Spontaneous macular hole closure with appearance of epiretinal membrane: implications for therapy. *Ophthalmic Surgery, Lasers Imaging Retin*. 2008; 39(3):237–238.
17. Shimada H, Nakashizuka H, Hattori T, Mori R, Mizutani Y, Yuzawa M. Double staining with brilliant blue G and double peeling for epiretinal membranes. *Ophthalmology*. 2009;116(7):1370–1376.
18. Sebag J, Wang MY, Nguyen D, Sadun AA. Vitreopapillary adhesion in macular diseases. *Trans Am Ophthalmol Soc*. 2009;107:35.
19. Michalewska Z, Michalewski J, Adelman RA, Nawrocki J. Inverted internal limiting membrane flap technique for large macular holes. *Ophthalmology*. 2010;117(10): 2018–2025.
20. Petropoulos IK, Matter MA, Desmangles PM. Spontaneous closure of macular hole: one-year follow-up with spectral-domain optical coherence tomography. *Klin Monbl Augenheilkd*. 2009;226(4):363–364.
21. Michalewska Z, Cisiecki S, Sikorski B, et al. Spontaneous closure of stage III and IV idiopathic full-thickness macular holes—a two-case report. *Graefes Arch Clin Exp Ophthalmol*. 2008;246(1):99–104.
22. Oh H. Idiopathic macular hole. *Microincision Vitr Surg*. 2014;54:150–158.