

Outcomes of vitreoretinal surgery in retinal detachment associated with morning glory disc anomaly

Parveen Sen, Puja Maitra, Harshit Vaidya, Pramod Bhende, Kalpita Das

Purpose: Morning glory disc anomaly (MGDA) is associated with a high prevalence of retinal detachment (RD), sometimes requiring multiple surgeries. The study aimed to establish the outcomes of RD surgery in such eyes and predict factors influencing the outcome, if any. **Methods:** It was a retrospective observational study of 9 eyes that underwent pars plana vitrectomy (PPV) for RD associated with MGDA. Clinical and surgical details were correlated with outcomes of surgery as noted at the final follow-up. **Results:** PPV was performed in 9 eyes. Lensectomy was done in 5 of 9 eyes (55.5%) during PPV. Laser photocoagulation around the disc was performed in 55.5% (5/9) of the eyes. Silicone oil tamponade was used in 77.7% (7/9) of eyes and gas tamponade (14% C3F8) was used in 22.3% (2/9) of eyes. Attached retina at final visit was observed in 66% of the eyes (6/9). Two out of 3 eyes that were seen to have preoperative glial tissue at the disc had poor outcome (odds ratio 10, $P = 0.16$). Five out of 7 (71%) eyes that had silicone oil tamponade, had an attached retina. No identifiable breaks were noted preoperatively in 5 eyes, of which 4 (80%) had an attached retina postoperatively. Vision improved by 1 line in 67% of the eyes that underwent surgery. **Conclusion:** MGDA related RD presents a unique set of challenges; meticulous PPV with or without lensectomy can help in achieving a successful anatomical and functional outcome in about two-thirds of patients.

Key words: Morning glory disc, morning glory disc anomaly, pediatric retinal detachment, retinal detachment in morning glory disc anomaly, retinal detachment surgery

Morning glory disc anomaly (MGDA) is a rare congenital nonprogressive, predominantly unilateral developmental defect of the optic nerve head (ONH) characterized by a large excavated optic disc surrounded by a ring of chorioretinal pigmentary disturbance, and radially oriented retinal vessels emanating from the disc giving it the appearance of a blossoming morning glory and believed to be a consequence of failure of fusion of neuro-ectoderm.^[1-3] Recently mesenchymal abnormalities resulting in incomplete closure of the posterior scleral wall and aplasia of the lamina cribrosa have been suggested as a possible cause.^[3] MGDA is considered a part of congenital cavitory optic disc anomalies.^[4] One-third of MGDA eyes are associated with retinal detachment (RD) which can follow an unpredictable clinical course of spontaneous attachment and re-detachment, sometimes requiring multiple surgeries.^[5,6] Management of retinal detachment poses a unique set of challenges during surgery. Despite anatomical success, visual prognosis is often guarded due to the inherent optic disc pathology and amblyopia. We aim to assess various aspects pertaining to the outcomes of surgical management of RD in MGDA.

Methods

All patients undergoing surgery for RD associated with MGDA from 2010 to 2019 at our tertiary care center were included. Nine eyes of 9 patients were included for retrospective analysis.

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Demographic details, clinical and imaging features, type of surgery and other surgical details, and anatomical as well as functional outcomes after a single vitreoretinal surgery were documented. All patients were evaluated by a pediatrician for associated systemic disorders. The study adhered to the tenets of declaration of Helsinki and institutional review board approval was obtained. This study was approved by the institution review board and ethics committee on 20.7.20.

Surgical technique

All eyes underwent a 3-port pars plana vitrectomy (PPV) with 23/25/27 G cutter and intravitreal triamcinolone acetonide (IVTA) was used at the surgeon's discretion to assist in posterior hyaloid separation. Additional steps were performed on a case-to-case basis. Encirclage of size 240 was placed in some eyes where complete vitreous base shaving was not possible. Laser photocoagulation using frequency doubled Nd-YAG with 532 nm or diode laser 810 nm was performed. Choice of tamponade (either with Silicone oil or C3F8) was decided by the operating surgeon.

Anatomical success was defined as attached retina at 1 month after surgery and at the final follow-up after a single retinal reattachment surgery. Retinal status was assessed at regular intervals. In eyes where silicone oil was used, it was

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considered for removal after a variable period depending on the status of the eye.

Statistical analysis

Descriptive statistics like mean, standard deviation, and median were obtained for factors like BCVA and follow-up period, and defined on continuous scale, while frequencies and percentage were used for categorical factors. The reviewed data was maintained using Microsoft Excel and analyzed using Microsoft excel 2013 (Microsoft Corp, Redmond, WA).

All the analyses were performed using SPSS version 14.0 (IBM Corp., Armonk USA). Odds ratio (OR) was calculated using binary logistic regression and frequencies were tested using the Chi-square test.

Results

Nine eyes of 9 patients with morning glory disc anomaly underwent PPV for retinal detachment. Details of each patient are tabulated in Table 1. The mean age at presentation with RD was 9.88+/-7.33 years (range: 2–24 years) with a male preponderance of 7:2. Seven cases had unilateral involvement with a normal disc in the fellow eye, whereas other 2 presented with bilateral RD associated with MGDA. Of these 2 cases, the fellow eye was inoperable in one patient and the other had undergone surgery elsewhere and was excluded from analysis.

Six eyes (66%) had total RD while 3 eyes (33%) had subtotal RD. Macular detachment was seen in all eyes. On indirect ophthalmoscopy (IDO) preoperatively, a retinal break was identified in only 1 eye. Optical coherence tomography (OCT) was performed preoperatively in 2 eyes; a break was noted on OCT, at the disc edge in one of these eyes [Patient no-1, Fig. 1]. In the other patient, though OCT failed to detect a retinal break, glial tissue at ONH was noted [Patient no-2, Fig 2]. The presence of glial tissue was seen on IDO in 2 other patients. Four eyes had proliferative vitreoretinopathy (PVR) changes at presentation in the form of subretinal gliotic bands (SRG).

Five of the nine eyes (55.5%) underwent lensectomy during PPV. An additional belt buckle (BB) was used in 6 cases to support the vitreous base. Five eyes (55%) had both lensectomy as well as BB. IVTA was used to aid in posterior vitreous detachment (PVD) induction in 55.5% (5/9) of the eyes. Retinal break was identified in 44% (4/9) of the eyes overall (preoperatively on IDO or on OCT, or intraoperatively). The location of these breaks noted preoperatively was confirmed intraoperatively. The retinal breaks were seen in close proximity to the anomalous disc in all 4 eyes (2 eyes had a break on the disc, 1 eye had a break in the inferotemporal edge of the disc, and 1 eye had a break in the papillomacular bundle). Iatrogenic break occurred in 2 eyes during PVD induction, and in both, the breaks were beyond the midperiphery. Subretinal fluid was drained through preexisting/iatrogenic breaks or through a planned retinotomy. The glial tissue over the disc was removed completely or trimmed to extent possible. Laser photocoagulation around the disc was performed in 55.5% (5/9) eyes. Silicone oil tamponade was used in 77.7% (7/9) of eyes and gas tamponade (14% C3F8/perfluoropropane) was used in 22.3% (2/9) of eyes. Preoperative and intraoperative details of each patient are tabulated in Table 1.

The mean follow-up was 18 months (1.5 months to 5 years) with a follow up of more than 1 year in 3 out of 9 patients.

Anatomical success in the form of attached retina was observed in 67% (6/9) eyes at the last follow-up. Three cases had total RD at the end of final follow-up. Eighty percent (4/5)

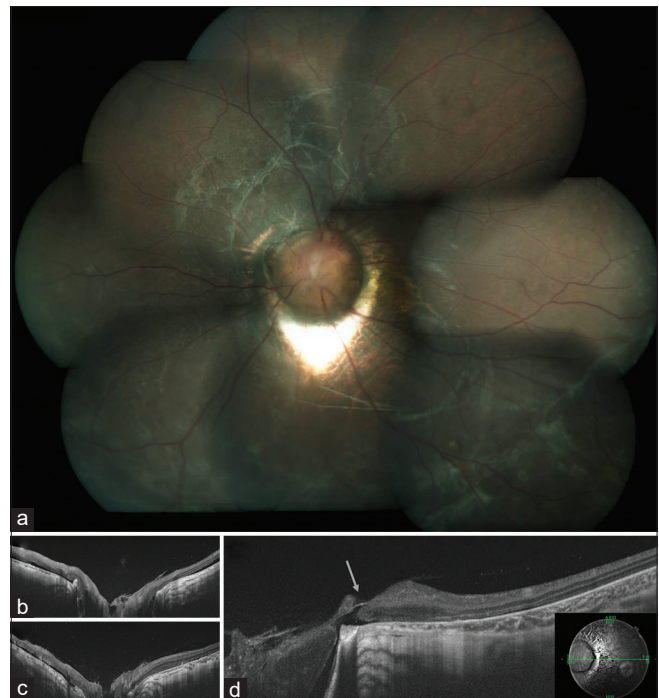


Figure 1: 6-year-old female (Case No. 1). (a) Subtotal retinal detachment of left eye with subretinal gliosis associated with morning glory disc anomaly. (b-d) OCT of the optic nerve head. Suspected area of break (white arrow) is localized on OCT preoperatively at the temporal disc edge

eyes with no identified breaks had an attached retina at final follow-up. Out of the eyes that had silicone oil as tamponade, anatomic success was seen in 71% (5/7), while 1 out of the 2 cases with gas tamponade showed anatomical success. One of the patients in present series (case 9) developed SRF over posterior pole within 2 months of surgery [Fig. 3], which was observed closely with postoperative serial OCTs, which did not detect any retinal breaks. The SRF resolved at 4 months follow-up [Fig. 3f].

Among eyes with silicone oil tamponade ($n = 7$), 3 eyes underwent Silicon oil removal (SOR). One of these eyes underwent lens aspiration with intraocular lens (IOL) implantation at the time of SOR. Of the other 4 eyes, 2 eyes had well-attached retina at last follow-up and 2 eyes had a recurrent retinal detachment. PVR changes in the form of subretinal gliosis (SRG) were seen preoperatively in 4 eyes of which 3 (75%) had attached retina at final follow-up. Two out of 3 eyes that had glial tissue at ONH had total RD at final follow-up.

Vision was improved by 1 line in 67% (6/9) eyes, maintained in 11% (1/9) eyes, and worsened by one or more line in 22% (2/9) eyes. Recurrent RD was the cause of drop in BCVA in both those eyes.

The presence of preoperative glial tissue over ONH appeared to be associated with a higher chance of unsuccessful anatomical outcome (odds ratio of 10), but the value was not statistically significant owing to the small number of cases in the series [OR 10.9, 95%CI 0.4–250, $P = 0.16$]. Presence of PVR, which in all cases was in form of SRG, did not appear to influence the anatomical outcome. Though significant associations with successful anatomic outcome could not be assessed because of a small sample size, absence of pre-existing breaks [OR 4.0, 95% CI 0.21–75.66, $P = 0.36$], and use of silicone oil as tamponade [OR 2.5, 95%CI 0.9–62.6, $P = 0.57$] appeared to be improving the final anatomic outcome [Table 2].

Table 1: Patient demographic details and findings

Eye	Age at RD (years)/ Gender	Complaints and findings	BCVA (log MAR)	Retinal detachment (RD)	Findings			Surgical details				Post op outcome			
					Preexisting Break	PVR	Other findings	BB	Laser around disc	Tamponade	Lensectomy	Drainage	Retinal Status	BCVA (log MAR)	
OS	6/F	Strabismus	1.3	Subtotal macula detached	Temporal edge of disc (noted on OCT and intraop)	SRG		no	yes	SiO	no		Through break	Attached (after SOR)	1.7
OD	13/M	DOV, Strabismus	1.3	Total RD	No	SRG	Glial tissue at ONH (seen clinically and on OCT)	no	yes	SiO	no		No retinotomy	Attached (after SOR)	1
OS	4/M	Leucocoria (cataract)	2.7	Total RD	At ONH (noted intraop after removing glial tissue)		Glial tissue at ONH (clinically)	yes	no	SiO	yes		Through iatrogenic break in SNQ	RD (oil filled)	2.3
OS	2/M	Leucocoria (cataract)	2.7	Total RD	No		Glial tissue at ONH (seen clinically)	yes	yes	C3F8	yes		Through drainage retinotomy in STQ	RD	2.3
OS	6/M	DOV, Strabismus	2.3	Subtotal RD macula detached	No			yes	no	C3F8	no		Through iatrogenic break at STQ midperiphery	Attached	2.3
OS	5/M	DOV, nystagmus	2.5	Total RD	No			yes	no	SiO	yes		Through Drainage retinotomy in SNQ	Attached (after SOR)	2.3
OD	8/M	DOV, nystagmus	2.1	Subtotal RD macula detached	At ONH (noted clinically on IDO)	SRG		yes	yes	SiO	no		Through break	RD (oil filled)	3
OS	24/F	DOV, strabismus	1.7	Total RD	PMB (noted at the time of vitrectomy)	SRG		yes	no	SiO	yes		Through iatrogenic at STQ midperiphery	Attached (oil filled)	1
OS	19/M	DOV, Strabismus	1.3	Total RD	No			yes	yes	SiO	yes		Through iatrogenic at ITQ	Attached (oil filled)	1

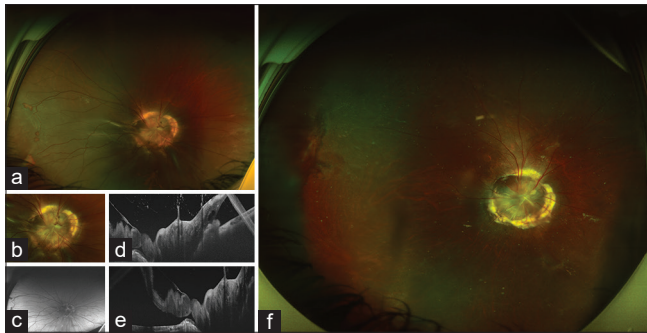


Figure 2: 13-year-old male (Case No. 2). (a) OPTOS image of the right eye with total retinal detachment associated with morning glory disc anomaly, also showing a wedge of peripheral avascularity and hemorrhages in the temporal periphery. (b) OPTOS image of the morning glory disc with evidence of glial tissue. (c) Red free image of the fundus of the right eye highlighting specs of hemorrhage temporally. (d and e) OCT of the optic nerve head revealing glial tissue which was noted clinically as well. (f) Postoperative images showing attached retina after silicone oil removal

Discussion

MGDA is commonly associated with RD in about 1/3rd cases. Surgical intervention with attention to the unique features seen in each case is necessary to salvage these eyes. Favorable anatomic outcome was seen in about 2/3rd of our cases and 78% maintained or improved vision at final follow up.

All three mechanisms of retinal detachment, viz., rhegmatogenous, tractional, and exudative, have been proposed.^[7-9] Retinal break may not be visible in all cases of RD as was the case in 55% of eyes in our series. The retinal breaks may be slit like and commonly located at the disc margin or within the anomalous optic disc where the retina is thin over an ectatic rim.^[7,8] Underlying choroidal tissue is atrophic resulting in loss of contrast for the breaks to be visible.^[7,8] OCT, especially 3D reconstruction image scan aids in better delineation of the disc anatomy, ascertaining the presence of epipapillary tissue or vitreous condensation and localization of breaks which may be caused by traction from epipapillary fibroglial tissue.^[8,10] We found OCT useful in these aspects, as well as in the follow up in a patient with persistent SRF postoperatively.

Lytvynchuk *et al.*^[9] used intraoperative OCT to support the tractional theory of RD in MGDA. They proposed that RD in early stages is usually tractional due to the epipapillary glial tissue. Unrelieved traction may lead to retinal break formation giving rise to a rhegmatogenous RD. Lytvynchuk^[9] and Matsumoto *et al.*'s^[11] hypothesis could explain why cases without obvious retinal break had twice as much chance of a successful outcome in our series. These cases were probably operated before formation of a "rhegma," which usually is at the disc margin and difficult to seal with any form of retinopexy.

MGDA can be associated with persistent fetal vasculature (PFV) of varying degrees suggesting a common underlying genetic abnormality involving the PAX6 (OMIM: 607108).^[12] Proliferative tuft of glial tissue seen over the ONH, now believed to be a spectrum in association with PFV, causes tangential traction thereby causing the retina to be bunched up around the disc margin.^[2,11,13] Chang *et al.*^[8] stated that the presence of glial tissue increased the probability of retinal detachment in MGDA and its removal or trimming remained vital to relieving traction.^[8,14] Three of the 9 eyes in our series revealed a glial tissue within the disc. It is firmly adherent and complete removal may not always be possible. Two out of 3

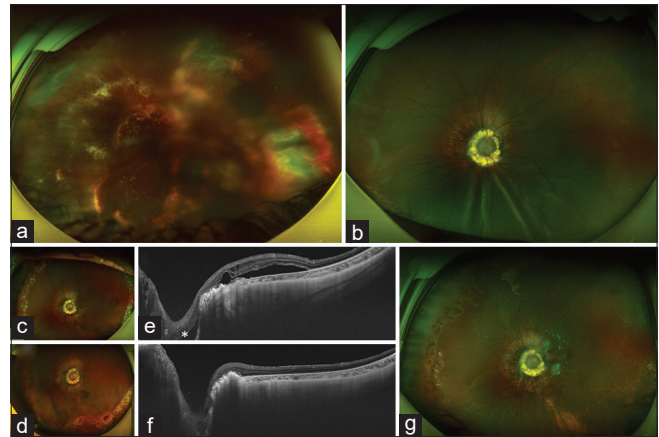


Figure 3: 19-year-old male (Case No. 9). (a) OPTOS image of chronic retinal detachment in right eye with no perception of light. (b) OPTOS image of left eye showed total retinal detachment associated with morning glory disc anomaly. (c and d) Early postoperative OPTOS image of the left eye showed shallow subretinal fluid over posterior pole and lasered peripheral break in inferotemporal quadrant. (e) Early postoperative OCT with shallow subretinal fluid at macula and cleft like space at the optic nerve (*). (f) OCT after 1 month of surgery showing resolution of subretinal fluid. (g) OPTOS image of the left eye after 1 month of surgery showing attached retina

eyes with glial tissue had a poor outcome in the form of total recurrent RD. This could be because of preexisting anatomical alterations, missed and untreated breaks at the base or at the proximity of the glial tissue, that are either formed due to traction caused while removing them or maybe preexisting breaks which get unplugged while trimming the glial tissue, ultimately causing detachment in eyes post vitrectomy. Removal of the internal limiting membrane (ILM) has been described to further relieve the tangential traction due to this glial tissue.^[9] ILM peeling in these cases may not always be possible because retina is thin and stretched, especially in the peripapillary area, and was not attempted in any of the cases in the present series.

The challenge of inducing posterior vitreous detachment (PVD) that is commonly seen in pediatric eyes, is particularly encountered in MGDA eyes with RD because of existence anomalous and strong vitreous condensation in peripapillary area.^[8] Four eyes in our series underwent IVTA assisted PVD induction. Reportedly MGDA eyes are associated with peripheral non perfusion and neovascularization akin to that found in familial exudative vitreoretinopathy (FEVR).^[15] Thus, one may come across anomalous vitreous adhesion and peripheral break formation intraoperatively or in the postoperative period [Fig. 3d]. Hence, these eyes may benefit from the use of an encircage as was used in nearly 77% of the eyes in the present study.

Another challenge is achieving an adequate retinopexy for retinal breaks at or within the ONH. Bartz-Schmidt and Heimann^[2] believed that laser around disc was not effective due to the absence of RPE around the disc for absorbing laser energy. Munk *et al.*^[16] using enhanced depth imaging OCT, however described the presence of a hyperreflective band extending into the anomalous disc that they believed to be the RPE. In these cases, diode laser is preferred as deeper penetration of his laser helps in preserving the nerve fiber layer around the disc margin.^[16] Laser photocoagulation around the disc margin in the absence of any obvious retinal breaks is controversial; it has the risk of damage to the nerve fibers at papillomacular bundle as well as the ONH. It was not done in a total of 4 cases. Out of these 4 cases, 3 had attached retina. We

Table 2: Assessment of factors determining the surgical outcome

Prognostic factors	Attached Retina (n=6)	Detached Retina (n=3)	Odds Ratio (95% CI)	P
Preexisting break identified				
Yes	2 (50%)	2 (50%)	4.0 (0.21, 75.66)	0.36
No	4 (80%)	1 (20%)		
PVR				
Yes	3 (75%)	1 (25%)	0.5 (0.03, 8.95)	0.64
No	3 (60%)	2 (40%)		
Glial tissue at ONH				
Yes	1 (33%)	2 (66%)	10.0 (0.40, 250.4)	0.16
No	5 (83.3%)	1 (16.7%)		
Tamponade				
C3F8	1 (50%)	1 (50%)	2.5 (0.9, 62.6)	0.57
Silicone Oil	5 (71.5%)	2 (28.6%)		
Vision at final follow-up (LogMAR)	1.47±0.64	2.63±0.35		0.03

PVR - Proliferative vitreoretinopathy; ONH - Optic nerve head

Table 3: Recent reports on RD associated with MGDA

Author	Journal/year	No of eyes	Details	Outcomes	Comments
Jo <i>et al.</i> ^[6]	Eur J Ophthalmol, 2011	1 Large hole in the excavated disc	Vitrectomy, laser around disc, and long-acting gas tamponade were done but had a re-detachment. A second operation using silicone oil tamponade was done	Retina was reattached after silicone oil-fluid exchange surgery.	Long standing tamponade implicated for success.
Chang <i>et al.</i> ^[14]	Eye, 2012	2 out of 6 eyes had RD associated with MGDA	Case 1: spontaneous resolution. Case 2: pneumoretinopexy with laser done - recurrence in 1 week. - vitrectomy with gas tamponade	Attached in all cases	In case 1: OCT aided localization of break in thin peripapillary retina. Retinoschisis in fellow eye
Fei <i>et al.</i> ^[12]	Br J O, 2013	8 eyes with RD 3 eyes operated.	2 eyes underwent C3F8 tamponade, 1 eye underwent 360 degree RR and SIO tamponade.	All operated eyes had attached retina	All eyes were associated with PFV (retrolental fibrous membrane/fibrous stalk)
Zhang <i>et al.</i> ^[21]	Eye Sci 2013	8 eyes Proliferative RD with hole in excavation of the optic disc	Vitrectomy with peripapillary laser and tamponade and removal of ERM and subretinal membrane with oil tamponade	Attached after SOR	
Lytvynchuk <i>et al.</i> ^[9]	BMC ophthalmology, 2017	1 eye (non rhegmatogenous RD)	Attached retina (23G, ERM/ILM peeling) with air endo tamponade	Attached	Intraoperative OCT assisted surgery
Ahuja <i>et al.</i> ^[20]	Niger J Ophthalmol, 2018	4 eyes	Triamcinolone-assisted vitrectomy, trimming of glial tissue, ILM peeling, laser along the excavated disc, SF6 (n=1) silicone oil (n=2) BB n=1	2 out of 3 eyes at attached retina (1 eye had re-detachment after SOR) and unsuccessful re-surgery	3 other eyes in the series had features of spontaneously attached RD
Jiang <i>et al.</i> ^[19]	BMC ophthalmology, 2019	1 eye (MGDA with PFV) no breaks	PPV + drainage retinotomy + laser around it + SIO tamponade	Attached retina	2 years follow up (oil filled eye)

MGDA - Morning glory disc anomaly, RD - Retinal detachment, PFV - Persistent fetal vasculature, OCT - Optical coherence tomography, ILM - Internal limiting membrane, ERM - Epiretinal membrane, SF6 - Sulfur hexafluoride, SIO - Silicone oil injection, SOR - Silicone oil removal, RR - Relaxing retinectomy

believe that either these cases had only a TRD to begin with or there was a small micro-break within the anomalous disc (not visible preoperatively or intraoperatively) and retina settled with vitrectomy and tamponade. TRD in young age group can be very high, mimicking a rhegmatogenous RD because of the

increased stretchability of retina seen in children as also seen in retinopathy of prematurity associated TRD. However, the absence of a detectable break within the anomalous disc on pre or intraoperative examination, does not rule out the presence of microbreaks.

In the absence of an obvious retinal break, various authors have proposed different sources for the subretinal fluid (SRF), including vitreous and cerebrospinal fluid (especially in young children, who neither have a liquified vitreous nor any posterior vitreous detachment), and leakage from retinal or choroidal vasculature.^[2,13,14,16,17] Recent imaging studies have elicited close proximity of subarachnoid space immediately posterior to the thin layer of tissue at the bottom of disc excavation in certain disc anomalies.^[16] Communication between subretinal space or vitreous cavity and subarachnoid space in eyes with MGDA has also been established by migration of dye and gas bubble.^[14,17,18] Due to this, there has been some concern regarding the use of silicone eyes as tamponade in these eyes with risk of migration of emulsified bubbles in subarachnoid space. Despite these concerns, Jo *et al.*^[16] and Bart-Schmitz and Heimann^[2] proposed the use of long-term tamponade like silicone oil as compared to gas.^[18] In our series, 70% of the eyes with good anatomical outcome had long-standing tamponade with silicone oil. Gas tamponade (C3F8 14%) was used in 2 of the 9 eyes. Though the sample size was small to establish a statistical significance, eyes with silicone oil tamponade had better outcome as compared with gas (70% vs 50%). Use of long-acting tamponade has been described by other authors as a favorable option especially in the pediatric age group and after recurrence^[6,9,12,14,19-21] [Table 3]. A close follow-up for signs of emulsification would be necessary to decide on early SOR. SOR was done in 3 eyes, and in one eye which developed significant cataract, SOR was combined with lens aspiration with IOL implantation. No re-detachment was noted after SOR until the last follow-up. None of our patients underwent re-surgery for RD. One eye had recurrence after C3F8 and was not operated due to advanced PVR; 2 eyes had recurrent RD under oil and had a thinned out retina. Re-surgery was not attempted due to anticipated poor anatomical and visual outcome.

Visual outcomes though not remarkable are important especially in patients where the other eye is inoperable or has extremely poor vision. Despite many challenges, surgical intervention is undertaken to provide or preserve ambulatory vision.

Limitation of the study is its retrospective design, small sample size, surgeon heterogeneity, and limited follow up. But given the rarity of disease and paucity of literature, we believe that our study will provide a useful update in the management of RD with MGDA.

Conclusion

In conclusion, keeping in mind the underlying mechanisms of RD in MGDA, management of these cases should include meticulous preoperative (preferably with OCT in all cases) and intraoperative examination to identify a retinal break, relieve maximal traction by removal of the epipapillary glial tissue, complete removal of the posterior hyaloid, and a long-standing endotamponade. RD in MGDA remains challenging; nonetheless, a careful PPV in experienced hands can help attain good anatomical outcomes and retain some functional or ambulatory vision.

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Conflicts of interest

There are no conflicts of interest.

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