Coloboma associated retinal detachment: Anatomical and functional results in the era of microincision vitrectomy surgery with an evaluation of risk factors for a recurrence

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Purpose: To analyze the anatomical and visual outcomes of microincision vitrectomy surgery (MIVS) with silicone oil tamponade in eyes having coloboma-related retinal detachment (RD) and evaluate the risk factors for recurrence of RD. Methods: This was a retrospective, multicentric analysis of eyes having coloboma RD undergoing MIVS with silicone oil tamponade between March 2010 and July 2018. Results: We evaluated 148 eyes of 144 patients. The mean age of presentation was 17.4 ± 9.8 years (range: 2–65 years) and the mean follow-up duration was 13.1 ± 13.8 months (range: 3–84 months). The single operation success rate was 88.5% (131 eyes), with an overall successful outcome achieved in 90.5% (134 eyes). Recurrence of RD occurred in 17 eyes (11.5%) over a mean duration of 2.59 ± 3 months. The risk of recurrence was found to be higher in eyes where relaxing retinectomy (RR) was performed (odds ratio [OR]: 3.22; \( P = 0.05 \)). A statistically significant improvement in vision was noted in the majority of cases from logMAR 1.85 ± 0.34 preoperatively to logMAR 1.33 ± 0.6 post-surgery (\( P = 0.002 \)). Conclusion: MIVS with silicone oil tamponade provided an anatomical success rate of 90.5% in eyes with coloboma RD with a significant improvement in visual acuity. Eyes in which RR was performed were susceptible to higher rates of re-detachment.

Key words: Coloboma, MIVS, retinal detachment, silicone oil, vitrectomy

Chorioretinal coloboma is a rare, congenital defect of the eye resulting due to incomplete closure of the embryonic fissure and is estimated to occur in 0.14% of the population.[9] The eyes with colobomatous malformation of the retina and choroid have a higher incidence (23–40%) of retinal detachment (RD) due to premature vitreous syneresis, inherent defects at the locus minoris resistentiae, and thin rudimentary retina with breaks involving the intercalary membrane (ICM).[10] The outcome of pars plana vitrectomy (PPV) in coloboma RD remains guarded owing to the following obstacles faced during surgery: difficulty in visualization secondary to the presence of microcornea, detection and closure of the breaks within the coloboma, and laser photocoagulation sparing the fovea at the edge of the coloboma. With the availability of wide-angle viewing systems and microincision vitrectomy surgery (MIVS), the outcomes in coloboma RD are expected to improve. We report our anatomical and functional outcomes of coloboma RD using MIVS. We additionally evaluated the risk factors for a re-detachment in these cases, which may further help in prognosticating the outcomes of this unique subset of retinal detachments.

Methods

A retrospective analysis of all consecutive patients of coloboma-related RD who underwent 23/25G PPV with silicone oil tamponade in the retina services of two tertiary eye care centers in North and Central India between March 2010 and July 2018 was performed. The study followed the principles of the Declaration of Helsinki and the institutional review board approval was obtained. The ethics committee approval was obtained on 15th October, 2020.

The hospital management system of both centers was used to draw the registration number of patients diagnosed with coloboma RD. All patients with a chorioretinal coloboma and rhegmatogenous retinal detachment (RRD) along with a coexisting ICM detachment with/without a peripheral break were included.

Patients with a primary peripheral break and RRD not extending within the coloboma, history of previous retinal...
surgery or laser, those having a follow-up of fewer than 3 months, or those cases with incomplete records were excluded.

The following pre-operative details were noted: demographic profile of patients, corrected distance visual acuity (CDVA), intraocular pressure (IOP), microcornea (corneal diameter <10 mm), nystagmus, iris coloboma, and lens status. The respective Amsler charts were reviewed to extract data regarding the type of chorioretinal coloboma (Ida Mann’s classification),[3] types of coloboma RD (Gopal et al.),[4] and the extent of RD. Intraoperative details included surgical steps performed, use of encircling band, site of drainage, and intraoperative iatrogenic breaks (IBs). Post-operative data included CDVA, IOP, and status of the retina at each visit. An IOP >21 mm Hg on three consecutive applanation tonometry readings was considered as raised.

The CDVA was measured using an electronic Snellen’s chart and was converted to logMAR for statistical analysis. For patients presenting with CDVA poorer than 3/60, visual acuity was evaluated as per the endophthalmitis vitrectomy study (EVOS) guidelines.[5]

Anatomical success was defined as a complete attachment of the retina outside the colobomatous area, with or without silicone oil in situ, at 3 months following surgery. Anatomical failure (recurrence) was defined as RD outside the coloboma area within 3 months of the primary surgery with or without oil in situ, or retinal detachment noted during silicone oil removal (SOR).

The risk for recurrence of RD was correlated with possible risk factors such as the type of coloboma, age <16 years, use of an encircling band, the performance of a relaxing retinectomy (RR), 360-degree laser barrage, fovea sparing laser, pars plana lensectomy (PPL), and the occurrence of an IB. The mean pre-operative and post-operative logMAR CDVA was compared to assess the visual outcomes. The primary outcome measure was the percentage of eyes with an attached retina (primary surgery success rate) at 3 months follow-up. The secondary outcomes measures included the visual outcomes and the risk factors for recurrence of RD.

Surgical details
All patients had undergone surgery performed by four senior (>10 years of experience in Vitreoretina) surgeons using standard 25G/25G MIVS with silicone oil tamponade. A #240 band (MIRA Inc., Waltham, MA, USA) was placed at the start of PPV based on the surgeon’s discretion. In all cases, the sclerotomy ports were placed at 4 mm from the limbus. This was done irrespective of the corneal diameter or lens status. In our experience, this helps in reducing the chances of the instrument touching the lens in such eyes with microcornea. The posterior vitreous detachment (PVD) was induced using active suction of a vitrectomy cutter. Triamcinolone acetonide was used for better visualization of the vitreous during PVD induction. A PPL was performed in patients with lens opacities obscuring the fundus view, accidental lens touch during vitrectomy, or in eyes with severe microcornea. A complete PPV with 360 degrees vitreous base dissection was performed using scleral indentation. Persistent posterior cortical vitreous was ruled out using triamcinolone acetonide. A thorough examination was done to identify any peripheral retinal break following which fluid air exchange (FAX) was performed either through a pre-existing retinal break or drainage retinotomy or breaks identified in the ICM. A posterior drainage retinotomy was made in cases wherein complete FAX could not be achieved via the ICM breaks and the retina ballooned at the edge of the coloboma. Three to four rows of endolaser photocoagulation was done along the entire margin of the coloboma using a frequency-doubled Nd: YAG laser (532 nm) or a diode red (810 nm) laser. In eyes where the fovea was within the coloboma or was abutting the coloboma margin, a single row of the laser was done or a fovea sparing laser was done. Circumferential laser retinopexy to the peripheral retina, including any pre-existing or iatrogenic break, was performed with 3–4 rows of the confluent laser, followed by 1,000–1,500 centistoke silicone oil (Aurosil®, Aurolab, Madurai, India) injection. RR was performed in select cases with anterior proliferative vitreoretinopathy (PVR) to ensure complete reattachment of the retina. The SOR was done once the complete retinal reattachment was achieved with good laser uptake and no evidence of PVR. The SOR was done using 23/25 gauge three-port PPV, following which the retinal attachment status was evaluated and a peripheral examination was performed intraoperatively. Eyes having emulsified silicone oil were subjected to additional fluid air exchange to facilitate oil droplet removal. Emulsification of silicone oil or silicone oil-induced glaucoma were other indications for SOR. The SOR was deferred in patients with a high risk of re-detachment. Patients were considered as having a high risk of re-detachment based on the duration of retinal detachment, the status of PVR, intraoperative assessment of the retina, postoperative evaluation, and presence of a large RR.

Statistical analysis
The data were entered in an MS Excel spreadsheet and analysis was done using Statistical Package for Social Sciences (SPSS) version 21.0. Categorical variables are presented in numbers and percentages, and continuous variables are presented as mean ± standard deviation (SD), and median. Normality of data was tested using the Kolmogorov–Smirnov test. If the normality was rejected, then a non-parametric test was used. Quantitative variables were compared using Wilcoxon rank-sum test for comparing CDVA. Qualitative variables were correlated using the Chi-square test. A P value of ≤ 0.05 was considered statistically significant.

Results
A total number of 148 eyes of 144 patients were included in the study, of which 88 (61.12%) were males and 56 (38.88%) were females. The mean age of presentation was 17.4 ± 9.8 years (range: 2–65 years) and a median of 16 years. The maximum number of patients (n = 68; 47.2%) was present in the second decade of life (interquartile range: 10–20). The right eye was operated on in 58 (38.19%) and the left eye in 90 (60.81%) patients.

The occurrence of other associated features along with chorioretinal coloboma and the type of coloboma RD is detailed in Table 1. Type 1 coloboma was present in 71 (48%) eyes, type 2 and type 3 were present in 38 (25.6%) and 39 (26.4%) eyes, respectively. The mean follow-up duration was 13.19 ± 13.85 months (range: 3–84 months).

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The details regarding surgical procedures are enumerated in Table 2.

IBs were found in 43 (29.05%) eyes during surgery. Out of these 43 eyes, breaks were adjacent to the peripheral edge of the coloboma in 30 (69.8%) eyes, whereas the rest 13 (30.2%) eyes had them in the superior peripheral quadrant. A single IB was noted in 26 (60.46%) eyes and 17 (39.54%) eyes had multiple IBs.

SOR was done for 97 (65.5%) out of 148 eyes; the mean time to SOR was 6.67 months (range: 1–60 months) (median: 4.5 months). The SOR was not advised in 41 eyes due to an increased risk of re-detachment considering the grade of PVR changes and 10 eyes were lost to follow up and were unable to undergo SOR.

The retina remained attached after the primary surgery (single operation success rate) in 131 eyes (88.5%) [Fig. 1]. Recurrence of RD occurred in 17 eyes (11.5%) over a mean duration of 2.59 ± 3 months. Twelve eyes had a recurrence after SOR, whereas five eyes had a recurrence with silicone oil in situ. Re-surgery was advised for all cases; however, only three patients underwent the same. In these three eyes, the retina was attached at the last follow-up under silicone oil. Therefore, the final anatomical success rate was 90.5%.

The odds ratios (ORs) of the risk factors for anatomical failure are detailed in Table 3. A significantly higher chance of recurrence was seen in eyes where RR was performed. Although eyes undergoing lensectomy, those with additional peripheral breaks, and those with type 1/2 coloboma had a higher risk for recurrence, the association was not statistically significant. Four out of 17 eyes that developed a re-detachment had multiple IBs during the primary surgery. The OR for the risk of re-detachment in eyes with multiple IBs was 7.7. The use of an encircling band, age <16 years, microcornea, fovea sparing laser, and the presence of a 360-degree laser barrage did not influence the outcomes in our series. Axial length is another important risk factor that may influence the rate of recurrence of RD. However, we were unable to accurately measure the axial length in this group of patients due to poor fixation.

The CDVA statistically improved from logMAR 1.85 ± 0.34 to logMAR 1.33 ± 0.6 (P = 0.002). Of the 148 eyes, 109 (73.6%) eyes showed an improvement in CDVA postoperatively. From the 119 eyes presenting with a CDVA of <1/60 (near blindness) (World Health Organization [WHO] classification), 60 eyes (50.4%) showed an improvement in the blindness grade.

An increase in IOP was seen in 37 eyes (25%), which ranged from 22 mm of Hg to 56 mm of Hg, with a mean of

### Table 1: Associated features along with chorioretinal coloboma and distribution of types of coloboma RD

| Associated ocular features with chorioretinal coloboma | Number of eyes (%) |
|-------------------------------------------------------|---------------------|
| Iris coloboma                                          | 57 (38.5)           |
| Lens coloboma                                          | 142 (95.9)          |
| Microcornea                                           | 90 (60.8)           |
| Nystagmus                                             | 54 (36.4)           |
| Squint                                                | 13 (8.7)            |
| None                                                  | 9 (6)               |

| Distribution of type of coloboma RD                     | Number of eyes (%) |
|--------------------------------------------------------|--------------------|
| Type II-B                                              | 75 (50.8)          |
| Type II-C                                              | 43 (29)            |
| Type II-D                                              | 0 (0)              |
| Type II-E                                              | 20 (20.2)          |

Table Legend: The first section of the table shows associated ocular features along with chorioretinal coloboma, whereas the second part shows the distribution of type of coloboma RD

### Table 2: Details of the surgical steps during pars plana vitrectomy

| Surgical procedures | Number of eyes (%) |
|---------------------|--------------------|
| Encircling band     | 128 (86.46)        |
| Lensectomy          | 108 (72.29)        |
| Relaxing retinectomy| 20 (13.51)         |

### Table Legend:
ICM: intercalary membrane, DR: drainage retinotomy

Figure 1: (a) Preoperative fundus photograph of a 17-year-old female patient with type 3 coloboma-associated retinal detachment and a BCVA of 5/60. (b) The 1-month postoperative fundus photograph shows an attached retina with a silicone oil reflex. The BCVA improved to 6/36. (c) The preoperative fundus photograph of a 16-year-old female patient with a type 3 coloboma-associated retinal detachment along with subretinal bands (white arrow). (d) One month postoperatively, the fundus picture shows an attached retina with a silicone oil reflex. The BCVA improved from hand motion at baseline to 6/60 postoperatively.
35.58 ± 9.51 mm of Hg. In 20 eyes (54%), the rise in IOP was noted within the first postoperative week, whereas the others had an IOP rise after a mean duration of 23 weeks (range: 2 to 104 weeks). The IOP was controlled with oral and topical antiglaucoma medications in 17 eyes, whereas in 15 eyes the increase in IOP was attributed to the silicone oil for which an early SOR was performed. Five eyes that were refractory to medical management or had persistent high IOP even after SOR, were treated with trabeculectomy (1 eye), glaucoma drainage device surgery (2 eyes), and cyclophotocoagulation (2 eyes). Intractable glaucoma with optic atrophy developed in five eyes despite medical and surgical intervention.

Discussion

The management of the eyes with coloboma RD remains both difficult to access and challenging to repair. The anatomical success rate in these eyes varies between 35% and 85%,[7] PPV with a long-acting tamponade such as silicone oil has shown to achieve better anatomical outcomes than scleral buckles in such eyes.[5,8] The final visual acuity depends on the involvement of the macula and disc by the coloboma.[9] The complicated nature of the RD in eyes with chorioretinal coloboma leads to a higher incidence of recurrence of detachment following the primary surgery. However, in most cases, a re-surgery for managing the recurrence has been shown to improve the final anatomical outcomes.[9]

Anatomical outcomes

The anatomical success rate of MIVS in coloboma RD in our series of patients was 90.5%, which was comparable to the success rate of PPV and oil tamponade reported in the literature.[8,10,11] However, it remains difficult to accurately compare the anatomical success rates between two studies due to the heterogeneity of the cases and differences in the definition of anatomical success. A review of the literature pertaining to the various studies on the surgical outcomes of coloboma retinal detachments is described in Table 4.

In our series, the risk of anatomical failure was higher in eyes requiring RR. Residual vitreous traction or intrinsic retinal shortening in eyes with an advanced grade of PVR necessitating a RR could be the reason for subsequent re-detachment, a risk
**Table 4: Summary of surgical procedures and anatomical success rates of studies evaluating coloboma-related retinal detachments**

| Authors with the year of publication | Sample size (eyes) | Gauge of vitrectomy (G)/tamponade | Anatomical success (%) | Risk factor(s) for recurrence | Complications |
|--------------------------------------|-------------------|----------------------------------|------------------------|------------------------------|---------------|
| Gopal et al.[13] 1991                | 17                | 20/silicone oil                  | 81.8%                  | Persistent secondary glaucoma (17.6%); band keratopathy (23.5%) |
| McDonald et al.[14] 1991             | 7                 | 20/gas                           | 100%                   | -                            | -             |
| Corcostegui et al.[16] 1992          | 7                 | 20/gas                           | 100%                   | -                            | -             |
| Gopal et al.[16] 1998                | 85                | 20/silicone oil or gas           | 81.2%                  | Severe PVR type 2E RD        | Raised IOP (18.8%) |
| Pal et al.[10] 2006                  | 42                | Not specified/silicone oil       | 88.1%                  | Raised IOP (11.9%); hazy media (16.6%); epithelial defect (16.6%) |
| Ramezani et al.[17] 2010             | 28                | 20/silicone oil or gas           | 92.9%                  | -                            | Cataract progression (100%); IOP rise (46.4%) |
| Nagpal et al.[9] 2012                | 46                | 20/silicone oil                  | 86.9%                  | PVR New breaks               | Raised IOP (10.8%); hazy media (4.3%); epithelial defect (4.3%) |
| Wei et al.[14] 2014                  | 5                 | 20/silicone oil                  | 100%                   | Raised IOP (40%); Band keratopathy (20%) |
| Abouammoh et al.[6] 2016             | 119               | 20 or 23/silicone oil or gas     | 87.4%                  | Cryotherapy                  | Iatrogenic sclerotomy-related breaks (5.9%); silicon oil migration into AC (3.4%); transient hypotony (3.4%); raised IOP (12.6%); cataract progression (5%) |
| Hocaoglu et al.[11] 2017             | 10                | 20 or 23/silicone oil           | 90%                    | -                            | Cataract (30%); raised IOP (10%); band keratopathy (10%) |
| Suwal et al.[12] 2019                | 13                | 23/silicone oil                  | 92.3%                  | -                            | Cataract (7.69%); raised IOP (30.7%); band keratopathy (7.69%) |
| Present study                        | 148               | 23 or 25/silicone oil           | 90.5%                  | Performance of RR           | Iatrogenic break (29.05%); raised IOP (25%) |

IOP: Intraocular pressure; AC: Anterior chamber

**Figure 2:** The table on the left indicates different categories of range of vision and their respective reading abilities. The bar diagram on the right shows the postoperative distribution of the visual acuity of 119 eyes, which had near blindness range of vision preoperatively. The first blue column indicates the number of eyes with near blindness range of vision preoperatively (119 eyes). The second column onward is the depiction of postoperative vision in different categories.
factor that has also been reported by Pal et al.[16] Performance of lensectomy, those with additional peripheral breaks, and those with type 1/2 coloboma had a higher risk for recurrence; however, the association was not statistically significant. Lensectomy is warranted to meticulously remove the anterior vitreous in eyes with microcornea and those with lens opacities. Its performance can reflect the fact that these eyes were smaller, with cataracts, and hence could have led to an increased recurrence due to inadequate visualization of the periphery. Even though eyes with coloboma RD have breaks in the ICM, tackling additional peripheral breaks remains important intraoperatively, which is reflected by the increased recurrences seen in these eyes in our series.

As the location of the fovea is frequently within or at the edge of the coloboma, there exists an incumbent risk of developing inadvertent foveal damage while applying laser burns to the coloboma margin.[18] Furthermore, the extension of retinopexy scars can also be responsible for the subsequent involvement of the fovea. Considering these factors, only a partial endolaser to the edge of the coloboma, sparing the foveal region, was performed in selected cases with type 1 and type 2 coloboma. Also, the functional border of the disc is spared when the coloboma involves the disc. This may have led to an increased risk of recurrence in these eyes.

IBs increase the risk of postoperative re-detachment if not adequately dealt with during the primary procedure. In our series, although a single IB was not associated with a higher risk of recurrence, multiple IBs increased the risk of re-detachment seven times. Furthermore, the IBs encountered in our series were most commonly located along the coloboma margin inferiorly. This can be explained by the strong vitreous adhesion and the shearing forces generated during PVD induction, straddling the transition zone between the thin atrophic, diaphanous ICM, and adjoining normal retina. This anomalous, strong vitreous adhesion has also been described histologically by Schubert.[19] Also, Liu et al.[20] noted that the ora serrata was located more posteriorly in colobomatous eyes. This factor has to be taken into consideration while inducing a PVD in these cases to avoid inadvertent breaks near the vitreous base, especially near the peripheral edges of the coloboma inferiorly.

Functional outcomes
The overall visual outcomes in our series were favorable with a significant gain in vision post-surgery. Despite a good anatomical reattachment of the retina, there was a compromise in acquiring optimal visual acuity in these cases due to the involvement of the disc and/or fovea in the colobomatous area.[10,13,16] However, in our series, even with the majority of eyes presenting with near blindness, half of the eyes attained an improvement in the blindness grade emphasizing the rewarding role of intervention in these cases.

A rise in IOP following vitrectomy was noted in one-fourth of our cases, of which 50% developed it within the first postoperative week. The early postoperative IOP surge has been partly attributed to postoperative inflammation and surgical trauma.[16] In continuum to the above, emulsification of the silicone oil with its anterior migration can contribute to late-onset of IOP rise.[16] In our series, a subset of eyes developed recalcitrant glaucoma not responding to the maximum medical therapy, subsequently developing glaucomatous optic atrophy despite the removal of the silicone oil. This can be explained by the pre-existing developmental and structural defects involving the angle in the eyes with colobomatous malformation and decompensation of the residual function following structural damage due to operative trauma and inflammatory stress.[21]

On review of the literature, the dataset of studies on coloboma RD involved multiple centers, multiple operating surgeons, and was mainly retrospective in design. Similarly, our study remains limited by the inherent drawbacks of a retrospective design along with multiple operating surgeons, lack of PVR grading, and a short follow-up. However, our study differs by reporting the outcomes in a large cohort of coloboma-related RD exclusively managed with MIVS, which is now the standard of care for vitreoretinal surgeries. We also analyzed the surgical complications and possible risk factors for re-detachment in these eyes.

Conclusion
To conclude, MIVS provides good anatomical and visual outcomes in coloboma RD, with a low rate of recurrence. The need for performing RR was associated with an increased risk of re-detachment. IOP elevation was a common postoperative complication predominantly developing within the first postoperative week.

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Conflicts of interest
There are no conflicts of interest.

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