Macular hole following successful stage 4B/stage 5 retinopathy of prematurity surgery

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To report three eyes surgically managed for stage 4B/stage 5 retinopathy of prematurity (ROP) with successful retinal reattachment resulting in macular hole formation after 2–3 months. One patient was observed, and two patients underwent membrane dissection and silicone oil injection and maintained attached retina at last follow-up. The case series is novel to present internal limiting membrane peeling around macular hole with hole closure in one patient which is hitherto not reported. Two of the three eyes received intravitreal bevacizumab injection for aggressive posterior ROP before surgical intervention.

Key words: Bevacizumab, lens-sparing vitrectomy, macular hole, retinopathy of prematurity

The surgical success rate of stage 5 and 4B retinopathy of prematurity (ROP) remains limited despite advancement in instrumentation.^[1-3] The macular drag, myopia, amblyopia, nystagmus, and late retinal detachment limit the functional success.^[4-9] Late retinal detachment can happen as a result of cicatricial changes following regressed ROP leading to retinal break formation, and most of these breaks present around the equator and some posterior to it.^[10] Macular holes are rarely seen following stage 4 or 5 ROP surgery and there are limited case reports on the same.^[10] We present a case series of three eyes which developed macular hole after successful retinal reattachment following a lens-sparing or lens-sacrificing vitrectomy for stage 4B and stage 5 ROP.

Case Reports

Case 1

A 1-month-old premature female baby born at 28 weeks of gestation with birth weight of 1300 g and hospitalization for 1 month for respiratory distress presented with aggressive posterior (AP) ROP and florid vascularization in both the eyes. The baby underwent laser photocoagulation to avascular

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retina in two sittings followed by intravitreal bevacizumab $(0.625 \ \mu g \text{ in } 0.025 \text{ mL})$ in both the eyes on separate sitting in view of florid extraretinal vascularization and traction; still the patient progressed to posterior pole tractional retinal detachment with fibrosis. The patient underwent lens-sparing vitrectomy in the left eye by 2 months of age. Traction relief was adequate after a 25-G bimanual surgery. Postoperatively retina attached but right eye progressed to total retinal detachment with unsuccessful retinal attachment after surgery. After 2 months, a large macular hole developed at the posterior pole in the left eye with localized retinal elevation [Fig. 1]. The patient underwent resurgery with bimanual membrane dissection, silicone oil injection, and laser augmentation. Internal limiting membrane (ILM) peeling or laser around the macular hole was not attempted. The patient is maintaining attached retina with type 2 closure, 6 months after surgery with medical management of raised intraocular pressure.

Case 2

A 2-month-old male baby born at 28 weeks of gestation with a birth weight of 1700 g with a month of neonatal intensive care monitoring for respiratory distress syndrome and neonatal jaundice along with septicaemia presented with stage 5 ROP in both the eyes. Lens-sacrificing vitrectomy was done in the right eye with 25-G bimanual dissection. The funnel opened and disc was visualized at the end of the surgery. The retina settled by 1 month with tortuous vessels and vascular looping. After 3 months, a large macular hole developed in the right eye [Fig. 2] with localized retinal elevation and it was observed.

Case 3

A 1-month-old female baby born at 30 weeks of gestation with birth weight of 1600 g and history of hospitalization for 20 days with respiratory distress and packed red blood cell transfusion for anemia presented with APROP with extraretinal vascularization and nondilating pupils. After laser in both the eyes, the patient underwent bevacizumab (0.625 mg in 0.025 mL) intravitreal injection. Despite aggressive laser, both the eyes progressed to stage 4B ROP. The flat fibrosis dragged the macula nasally in both the eyes. The child required further blood transfusion to correct the anemia and the surgery was done after a month. Lens-sparing vitrectomy with 25 G was done in the left eye first. Vitreous was adherent at fovea and arcades with nasal drag toward the disc. Traction was relieved with careful scissors dissection. After 2 months, a macular hole was noted with shallow detachment of retina at the posterior pole. The patient underwent resurgery with bimanual dissection of posterior vitreous from the retina and ILM peeling after Brilliant Blue staining followed by silicone oil

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Figure 1: Case 1, (a) colour fundus picture of left eye with stage 4 B ROP. Intraoperative picture of left eye after adequate traction relief (b), postoperative picture of the left eye with attached retina at 1 month (c), and intraoperative picture of left eye with macular hole and localized retinal detachment (d)



Figure 3: Case 3 at presentation (a), fundus picture of left eye reveals stage 4B ROP. (b) After lens sparing vitrectomy (c) At 8 weeks followup with a macular hole with localised retinal detachment. (d) Resurgery included ILM peeling after Brilliant Blue staining. (e) Silicone oil injection. (f) At the last follow-up, the retina remained stable with closed macular hole

injection [Fig. 3]. ILM was densely adherent to the retina and difficult to peel; still a small 2-disc diameter area was peeled. At last follow-up after 4 months, the retina was stable with closed macular hole.

Discussion

Macular hole is a rare occurrence in patients operated for stage 4 or stage 5 ROP although retinal breaks as late complication of ROP sequelae/post ROP surgery have been reported. The present series reports occurrence of macular hole 2–3 months following surgery for stage 4 and stage 5 ROP. There is a single publication by Ahmad and Hirose, reporting macular hole with RD and the subsequent surgery for macular hole in four patients operated for stage 4 or stage 5 ROP. The maturity (28- to 30-week gestation) and birth weight (1300–1700 g) of the babies in the present series is better compared with the reported series with 24- to 28-week gestational age and 622–1200 g birth weight.^[10]



Figure 2: Case 2, (a) with stage 5 ROP and retina behind the lens. Intraoperative picture (b), of lens-sacrificing vitrectomy. Fundus picture (c), at the end of surgery with opened funnel and visualization of optic disc. At 3 months of follow-up (d), attached retina with a large macular hole

requiring anti-vascular endothelial growth factor injections and babies with eventful postnatal course. The present series also differs with early development of macular hole compared with the published report where macular hole developed after 15-33 months. Bevacizumab and laser would have accelerated the fibrosis in APROP cases resulting in increased traction posteriorly in the present series. The surgery for ROP does not aim to induce posterior vitreous detachment (PVD), and PVD may occur later and cause a tangential traction leading to formation of macular hole even after relief of anteroposterior traction and the tangential traction exerted by the ridge after a successful stage 4/5 surgery. Posterior ROP has stiffness of retina at the arcades leading to possible foreshortening, and when the retina falls back after traction relief it leads to tangential traction at the center with resultant hole at fovea as it is the thinnest part of retina; a similar observation was noted by Ahmad and Hirose in one patient where the macular hole enlarged following fluid-air exchange and they recommended macular bucking for such patients.^[10] The series is unique as it describes for the first time a successful closure of macular hole following ILM peeling in an ROP kid. The previous series described macular buckle in three patients for closure of macular hole.

Conclusion

One needs to be cautious and follow-up stage 4 and 5 ROP patients closely as macular hole can develop in such eyes any time postoperatively, and more severe posterior disease along with bevacizumab may further increase the risk. Surgery with good traction relief and ILM peel may help in closure of the hole.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflicts of interest

There are no conflicts of interest.

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