Case Report

Surgical Approaches to Optic Disc Pit Maculopathy: A Clinical Case Series

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Keywords
Optic disc pit · Maculopathy · Optic nerve head · Vitrectomy · Vitreoretinal surgery

Abstract
The purpose of this study was to compare the clinical outcomes of 13 patients with optic disc pit maculopathy (ODP-M) – progressive visual loss, serous macular detachment, and/or intraretinal fluid – who underwent different surgical approaches. This was a retrospective study including a consecutive sample of 13 patients aged 13–74 years (mean 35.38 ± 19.66 years) diagnosed with ODP-M and submitted to vitreoretinal surgery between 2005 and 2021. All patients underwent pars plana vitrectomy, posterior hyaloid detachment, and gas tamponade. Endolaser photocoagulation was applied to the temporal margin of the optic disc in 8 cases; internal limiting membrane (ILM) peeling was performed in 9 cases; and ILM inverted flap technique in 5 cases. Stuffing of the pit with an ILM flap was performed in 3 cases. Mean best-corrected visual acuity improved from 20/200 (1.04 ± 0.56 LogMAR) to 20/50 (0.43 ± 0.54 LogMAR) within 4–36 months. Central retinal thickness decreased from 587.5 ± 158.01 μm to...
253.9 ± 33.55 μm, and 7 out of 10 patients had complete resolution of intraretinal fluid. All patients had complete retinal reattachment; however, a few years after surgery, 4 patients had recurrence of serous retinal detachment. The only adjunctive technique associated with greater visual improvement was endolaser (p = 0.033) and not performing peeling of the ILM was also associated with better visual results (p = 0.013), independently of preoperative visual acuity or age at the time of surgery. None of the adjunctive procedures was a significant predictor of better anatomical outcomes. In conclusion, all of these approaches for the surgical management of ODP-M were safe and effective. In this study, vitrectomy with endolaser was a good option for management of ODP-M.

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Introduction

Optic disc pit is a congenital malformation of the optic nerve head that can lead to serous macular detachment, intraretinal and subretinal fluid accumulation, and macular schisis, consisting of optic disc pit maculopathy (ODP-M) that causes progressive visual impairment [1, 2]. Pars plana vitrectomy (PPV) is commonly employed for the treatment of ODP-M, but there are many adjunctive techniques such as endolaser photocoagulation, internal limiting membrane (ILM) peeling with or without inverted ILM-flap technique, and ILM stuffing of the pit, whose added benefit is still unclear to date [1–4]. We performed a retrospective study on 14 consecutive surgical cases of ODP-M to describe and identify the adjuvant techniques leading to improved outcomes.

Methods

This was a retrospective, interventional study of 13 eyes from 13 patients with ODP-M, who underwent vitreoretinal surgery at the Centro Hospitalar e Universitário de Coimbra between 2005 and 2021. Clinical data were gathered by the first author by consulting medical records and all patients were anonymized. Inclusion criteria were (1) a diagnosis of ODP, confirmed through fundoscopy (a unilateral and usually temporally-located greyish oval depression of the optic disc) and macular optical coherence tomography (OCT) analysis (showing communication between the optic nerve and the subretinal space); (2) signs or symptoms of progressive loss of visual acuity (VA) in the eye with the ODP with no improvement within 3 months; and (3) evidence of ODP-M, including macular detachment on fundoscopy, subretinal or intraretinal fluid (IRF) or macular schisis on OCT. All surgeries were performed by two different surgeons (F.H. and J.F.). Spectralis® (Heidelberg Engineering, Heidelberg, Germany) SD-OCT was used to evaluate the central retinal thickness (CRT), macular detachment due to subretinal and IRF in most patients, and Zeiss Cirrus 5000 HD-OCT® (Zeiss Meditec Inc., Germany) in 2 patients. We compared CRT, macular detachment, and IRF only in patients submitted for preoperative and postoperative evaluation by Spectralis®. All patients underwent vision assessment, fundoscopy, and OCT in the first month after surgery, then monthly during the first year, and those with a favourable course returned once every 2 months for the following year. Measurements of VA were recorded in ETDRS letter score notation and converted to LogMAR for statistical analysis. Reoperations in the same patient were excluded from statistical tests but were included in the descriptive statistics. The data were analysed using SPSS for Windows (v. 25.0 SPSS) and included Mann-Whitney U tests and multivariable regression models adjusting for selected confounding variables to determine the association between each surgical technique and best-corrected visual acuity (BCVA) improvement.
Results

The study included 14 surgeries of 13 patients, of which 9 were women. Patients were aged between 13 and 76 years (mean 35.38 ± 19.66 years). Surgery was performed within 3–6 months of the onset of symptoms, except for patients 4, 8, and 9, who presented with signs of long-standing macular detachment on OCT (significant distortion or hyperreflectivity of retinal layers and large macular detachments) and patient 10, who had clinical fluctuation of vision and of the macular detachment prior to surgery. Mean preoperative BCVA was 20/200 (1.04 ± 0.56 LogMAR). Mean preoperative CRT was 587.5 ± 158.01 μm, all patients had serous retinal detachment, and 10 (71.4%) patients had IRF. All patients were phakic except for patient 7, who was pseudophakic; patient 4 was submitted to cataract surgery. All patients underwent 23-Gauge or 25-Gauge PPV and posterior vitreous detachment (PVD) induction, assisted by triamcinolone acetonide. Low-fluence argon laser was applied on the temporal disc margin in 8 cases. The ILM was peeled in 9 cases, and an inverted flap of ILM was used to cover the pit in 5 patients (Fig. 1). Stuffing of the pit with ILM was executed in 3 cases. Fluid-air exchange using a silicone-tipped extrusion cannula was routinely performed, followed by gas tamponade with either 14% perfluoropropane (C3F8) (n = 3) or 20% sulphur hexafluoride (SF6) (n = 11). These results are summarized in Table 1.

Follow-up time varied between 1 and 16 years. Vision improved in all but 2 patients, mean postoperative BCVA was 20/50 (0.43 ± 0.54 LogMAR), and the time to achieve BCVA varied between 4 and 36 months (mean 15.43 ± 10.64 months). Resolution of macular detachment was achieved in all patients; however, macular schisis was still visible on OCT in...
Table 1. Summarized description of demographic details of the 13 patients with optic disc pit maculopathy, preoperative parameters, surgical procedures, and postoperative outcomes

| Patients | Age at diagnosis | Age at surgery | Gender | Duration of symptoms (months) | Endolaser ILM peeling | ILM flap | ILM stuffing | Preop BCVA | Postop BCVA | Months to achieve BCVA | Preop CRT, μm | Postop CRT, μm | Preop IRF | Postop IRF | Years until recurrence | Follow-up years |
|----------|------------------|----------------|--------|-------------------------------|------------------------|---------|-------------|------------|------------|-------------------------|--------------|--------------|----------|-----------|-------------------------|-----------------|
| 1        | 12               | 14             | M      | 4                             | Yes                    | No      | No          | 20/400     | 20/20      | 36                       | –             | –            | Yes       | No        | –                       | 11              |
| 2        | 38               | 42             | F      | 5                             | Yes²                   | No      | No          | 20/63      | 20/63      | 7                        | 418           | 277          | Yes       | No        | –                       | 8               |
| 3        | 19               | 20             | F      | 4                             | Yes                    | Yes     | No          | 20/63      | 20/40      | 4                        | 499           | 216          | Yes       | Yes       | 5                       | 6               |
| 4        | 54               | 54             | F      | 10                            | No                     | Yes     | Yes         | CF         | CF         | –                       | 582           | 227          | No        | No        | 3                       | 4               |
| 5        | 11               | 13             | F      | 3                             | No                     | Yes     | No          | 20/50      | 20/25      | 3                        | 472           | 278          | Yes       | No        | –                       | 5               |
| 6        | 39               | 39             | F      | 2                             | Yes                    | Yes     | Yes         | 20/63      | 20/25      | 12                       | 890           | 280          | Yes       | Yes       | –                       | 1               |
| 7        | 70               | 76             | M      | 6                             | Yes                    | Yes     | Yes         | 20/200     | 20/25      | 22                       | 570           | 307          | Yes       | No        | –                       | 4               |
| 8        | 51               | 52             | F      | 14                            | Yes                    | No      | No          | 20/400     | 20/63      | 12                       | –             | –            | No        | No        | –                       | 13              |
| 9        | 55               | 55             | M      | >24                           | No                     | Yes     | No          | CF         | 20/400     | 36                       | –             | –            | Yes       | No        | 3                       | 9               |
| 10       | 33               | 34             | M      | 8                             | Yes                    | No      | No          | CF         | 20/50      | 12                       | –             | –            | Yes       | Yes       | –                       | 16              |
| 11       | 25               | 25             | F      | 2                             | Yes                    | Yes     | No          | 20/63      | 20/32      | 12                       | 603           | 261          | Yes       | No        | –                       | 4               |
| 12       | 20               | 21             | M      | 2                             | Yes                    | No      | No          | 20/100     | 20/25      | 18                       | 469           | 247          | Yes       | No        | –                       | 7               |
| 13       | 15               | 15             | M      | 3                             | No                     | Yes     | No          | 20/63      | 20/25      | 12                       | 529           | 249          | No        | No        | –                       | 4               |

BCVA, best-corrected visual acuity; CF, counting fingers; CRT, central retinal thickness; ILM, internal limiting membrane; IRF, intraretinal fluid; Preop, preoperative; Postop, postoperative.

¹Reoperation on the first patient 11 years after the first surgery.

²Postoperatively.
one case (patient 6). Four patients had recurrence of serous macular detachment 3, 5, and 11 years following surgery; out of these 4 patients, 2 improved spontaneously, one underwent reoperation with completion of the PVD and ILM stuffing technique, and the other patient died in the meantime. There were no surgical complications except for a cortical cataract in patient 3, 6 months following surgery.

All except for 2 patients achieved a BCVA equal to or better than 20/80 irrespective of surgical technique. Endolaser was associated with a greater visual improvement \( p = 0.037 \), even after adjusting for preoperative BCVA \( p = 0.03 \) and age at the time of surgery \( p = 0.049 \). The mean BCVA improvement in eyes with no endolaser was 13.40 ± 7.76 ETDRS letters \( (-0.31 \pm 0.2 \text{ logMAR}) \) and 38.38 ± 19.04 ETDRS letters \( (-0.79 \pm 0.42 \text{ logMAR}) \) in eyes that were submitted to endolaser \( p = 0.033 \). Patients who did not undergo ILM peeling had also better visual outcomes (mean BCVA improvement 46.4 ± 16.04 ETDRS letters \[-0.97 \pm 0.38 \text{ logMAR}\] versus 17.75 ± 12.78 ETDRS letters \[-0.38 \pm 0.26 \text{ logMAR}\], \( p = 0.013 \). In regression analysis, not peeling the ILM was associated with greater visual improvement \( p = 0.007 \), even after controlling for preoperative BCVA \( p = 0.02 \) and age at the time of surgery \( p = 0.008 \). Neither endolaser nor peeling the ILM influenced the time required to achieve the BCVA \( p > 0.05 \). None of the other surgical techniques – ILM flap over the pit, ILM stuffing and C3F8 or SF6 tamponade – were significant predictors of BCVA improvement \( p > 0.05 \).

Regarding anatomical parameters, mean CRT improved from 587.5 ± 158.01 μm to 253.9 ± 33.55 μm, all patients had complete resolution of serous macular detachment and 7 out of 10 patients had complete resolution of IRF. None of the adjunctive techniques herein described was a significant predictor of postoperative CRT or CRT improvement, resolution of macular detachment or IRF \( p > 0.05 \).

**Discussion**

In this case series of 13 patients with ODP-M submitted to different surgical techniques, most patients had visual improvement within 3 years of surgery and these outcomes were sustained over time. We found that patients with signs of ODP-M chronicity (patients 4, 8, and 9) had worse postoperative BCVA, but also, patients with worse preoperative BCVA had better visual improvement, in accordance with other findings [4–6]. Despite some case series describing spontaneous resolution of the OPM [7, 8], early surgical intervention can prevent prolonged damage to the photoreceptors [4, 9].

Endolaser photocoagulation can theoretically create a scar that will act as a barrier to communication between the pit and the inner retinal layers and thus prevent leakage of fluid to the macula [10, 11], and from these results, endolaser led to significantly better visual outcomes. Nevertheless, this procedure is not without risks, and some prior studies concluded that endolaser provided no additional benefit for ODP-M [12–14]. The ILM might exert tangential force on the retina and contribute to the maculopathy, and its removal ensures hyaloid detachment in the macular area [13]. In this analysis, however, patients who did not undergo ILM peeling had better visual outcomes independently of preoperative BCVA or age at surgery (Table 1). Other authors have reached the same conclusions [14–16], and both the 2014 EVRS Optic Pit Study [17] and a recent review [3] do not support the need for ILM peeling.

The ILM flap and ILM stuffing techniques provide a physical barrier against the flow of fluid [5], and they can also act as a scaffold for the proliferation of Müller cells and consequential gliosis, further contributing to the barrier [10]. Previous comparison studies have shown that ILM stuffing of the pit had better or faster surgical outcomes than ILM peeling alone [18–20]. However, neither the ILM flap nor ILM stuffing influenced VA improvement in this study, which is in line with two recent reviews [3, 4]. Regarding gas
endotamponade, there were no differences in VA improvement between the SF6 and C3F8 groups, and other reviews highlight only the importance of gas tamponade following vitrectomy, as the gas acts as a temporary barrier between the vitreous cavity and the ODP and also pushes the intraretinal fluid into the outer layer of detachment, facilitating reattachment of the retina, regardless of the type of gas used [1, 14].

All patients had complete macular reattachment and a reduction of CRT followingsurgery. None of the adjunctive procedures were significant predictors of resolution of macular detachment, IRF or CRT improvement, in line with other studies [4, 12, 15]. Some authors claim that even though endolaser and ILM peeling are associated with a faster anatomical result, these are not necessarily associated with better visual outcomes [10, 18].

Four patients had recurrence of macular detachment and worsening vision; two of them recovered spontaneously, and one underwent reoperation with more adjunctive procedures, including removal of hyaloid remnants of the posterior pole and ILM stuffing. This has been recommended by Meng et al. [1], who stated that additional manoeuvres besides PPV should be used with caution in the first surgery.

In conclusion, there is a wide array of surgical procedures for the treatment of ODP-M, but no standardized evidence-based protocol is available. In this case series of surgical management of ODP-M, most patients achieved good long-lasting visual and anatomical results, regardless of the surgical technique. Endolaser photocoagulation and not peeling the ILM were the only significant predictors of better visual outcomes. According to our results, vitrectomy with endolaser and gas tamponade is a good option for the management of ODP-M.

**Statement of Ethics**

Ethical approval is not required for this study in accordance with local or national guidelines. The study followed the tenets of the Declaration of Helsinki and written informed consent was obtained from the patients for publication of these case reports and accompanying images and all data were anonymized.

**Conflict of Interest Statement**

The authors have no relevant financial or non-financial interests to disclose.

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**Author Contributions**

Rosa Lomelino Pinheiro: first author, data collection, and manuscript elaboration. Filipe Henriques: surgeon, project supervisor, project conception, and manuscript revision. João Figueira: surgeon and manuscript revision. Mário Alfaia: manuscript revision and Head of the Retina-Vitreous Department of the Centro Hospitalar e Universitário de Coimbra. Joaquim Neto Murta: manuscript revision and Head of the Ophthalmology Department of the Centro Hospitalar e Universitário de Coimbra.
Data Availability Statement

All data generated or analysed during this study are included in this article. Further enquiries can be directed to the corresponding author.

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