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Combined Approach for Management of a Case of Peripapillary Choroidal Hemangioma

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Abstract
Circumscribed choroidal hemangioma (CCH) is a rare vascular benign tumour of the choroid that typically presents as a solitary red-orange mass within the posterior pole in middle-aged adults with clinical symptoms of progressive vision loss, floaters, metamorphopsia and field defects. Despite its benign nature, it can present with complications, including subretinal fluid (SRF), serous detachment, cystoid macular oedema, and, in chronic cases, retinoschisis, retinal pigment epithelium changes, and neovascular glaucoma. Choroidal metastasis and choroidal melanoma have a close resemblance to choroidal haemangioma, which leads to a diagnostic dilemma. Imaging tools such as ultrasonography, optical coherence tomography, optical coherence tomography angiography, fluorescein angiography and indocyanine green angiography aid in the diagnosis making. Treatment aims to decrease exudation and thereby improve visual acuity in symptomatic patients. Photocoagulation, radiation therapy, photodynamic therapy (PDT), transpupillary thermotherapy (TTT), oral beta blockers and anti-vascular endothelial growth factor (VEGF) therapy are the available treatment options. In our case, we report a patient of circumscribed peripapillary choroidal hemangioma managed with a combined approach-based treatment using intravitreal anti-VEGF and oral beta blocker therapy. Due to its close proximity to the optic nerve of CCH, its treatment poses a challenge. Our case illustrates how a combined approach facilitates the successful management of the lesion.

Keywords: Circumscribed peripapillary choroidal hemangioma, vascular benign tumour, serous detachment, intravitreal anti-vascular endothelial growth factor, oral beta blocker

Introduction
The choroidal haemangioma (CH) is a rare vascular tumour of the choroid, which is benign in nature and can occur in two forms: circumscribed or diffuse. The circumscribed choroidal haemangioma (CCH) is usually sporadic, common in the fourth to sixth decades, and does not have any systemic manifestations [1-3]. It has a high incidence in the Caucasian population, particularly among males [1]. CCHs commonly involve the posterior pole, mainly in the super-temporal quadrant close to the macula [4, 5]. Histologically, the choroidal haemangiomas can be of capillary, cavernous or mixed type of blood vessels [5]. Usually, the CCH is the cavernous one of them. The diagnosis is made incidentally during a fundus examination, or it can be secondary to visual symptoms like blurred vision, floaters, metamorphopsia, and field defects. Hyperopic shift and secondary refractive amblyopia can occur in sub-foveal CCH. In more than half of the cases, complications such as subretinal fluid, retinoschisis, serous retinal detachment, secondary retinal pigment epithelium changes, and neovascular glaucoma occur, leading to a decrease in visual acuity (VA) of 20/200 or worse [1].
Close similarity to other choroidal tumours, such as choroidal melanoma and choroidal metastasis, makes it important to differentiate between them. Imaging tools such as ultrasonography, fluorescein angiography (FA), indocyanine green angiography (ICGA), magnetic resonance imaging (MRI), and optical coherence tomography (OCT) aid in the diagnosis [1, 6]. Photocoagulation, TTT, radiation therapy, PDT, anti-VEGF therapy and oral beta blockers are the available treatment options. Considering the peripapillary location, the minimally invasive combined approach of intravitreal anti-VEGF with oral beta-blockers is a step forward for management.

Case report

A 43-year-old female from Lucknow, Uttar Pradesh, presented with the chief complaint of blurring of vision in her left eye for three years with no systemic history. A thorough clinical examination was done, including fundoscopy, fundus fluorescein angiography and B-Scan for confirmation of the diagnosis of choroidal haemangioma. Best corrected visual acuity (BCVA) was 6/6 right eye (RE) and 5/60 left eye (LE). The Amsler grid showed left-sided metamorphopsia. On fundus examination, a 2.5DD orange-red elevated lesion nasal to disc (fig. 1a) with serous retinal detachment was noted.

Fig 1: (a) Fundus photo showing peripapillary circumscribed choroidal hemangioma (CCH) presentation. (b) Fundus photo of CCH at 8 weeks after management (c) Fundus photo of CCH at 16 weeks of follow-up

Spectral domain optical coherence tomography (SD-OCT) and OCT Angiography demonstrated a large choroidal mass with surrounding sub-retinal fluid, and the macula showed serous detachment (fig. 2a).

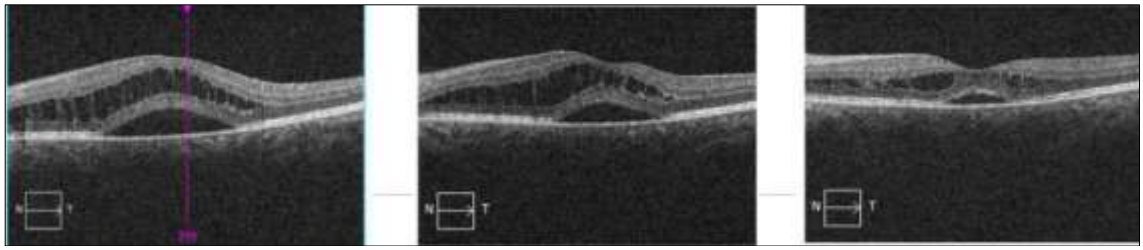

Fig 2: a) OCT showing subretinal fluid (SRF) at presentation, b) Decreased SRF on OCT at 8 weeks, c) Minimal SRF at 16 weeks of follow-up

B-scan showed a hyperechogenic dome-shaped mass ~ 6.6 x 3 mm (fig. 3a) with high internal vascularity, with subretinal fluid collection ~ 4.3 x 2 mm on the poster nasal quadrant of the LE. On fluorescein angiography, hyperfluorescence was seen (fig. 3b). Diagnosis of peripapillary circumscribed choroidal haemangioma (CCH) LE was made.

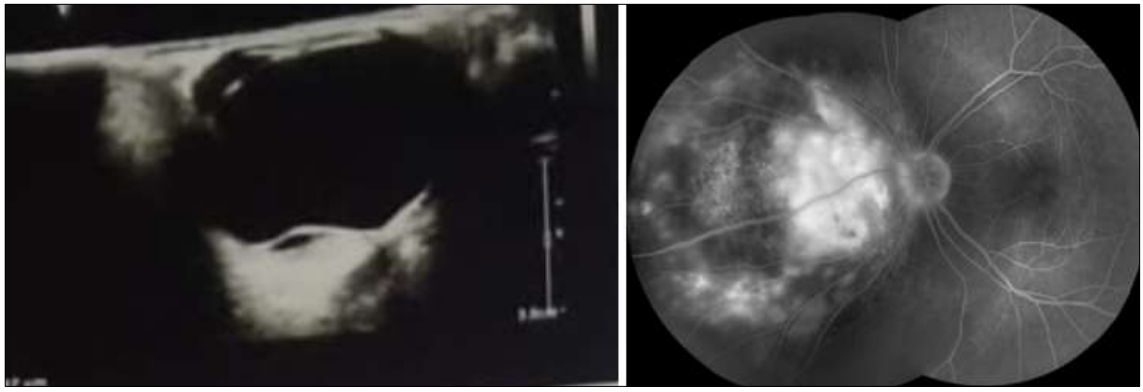

Fig 3: a) B-Scan showing echogenic dome-shaped mass ~ 6.6 x 3 mm in the poster-nasal quadrant of the left eye, b) Fundus fluorescein angiography showing hyperfluorescence

Treatment/Management

Medical Management: The patient was started on 10mg of propranolol, administered three times daily. 8 weeks later, the dose was increased to 20mg thrice daily.

Surgical Management: First dose of intravitreal injection (Ranibizumab) in the LE was also given.
A repeat dose of the same injection was given 8 weeks later.

Results: Follow-up was done at regular intervals. The patient improved symptomatically with BCVA at 8 weeks, 6/36, and at 16 weeks, 6/18, in the LE. Marked improvement was seen on fundus and OCT examination at 8weeks (fig. 2b & fig. 3b respectively) and 16weeks (fig. 2c & fig. 3c respectively). The patient had been planned for laser photocoagulation around the nasal half once the fluid receded.

Discussion

Asymptomatic patients do not require any treatment, but close periodic follow-up is needed. Conversely, in symptomatic individuals, the treatment depends on tumour size and location, the presence of SRF and potential visual gain. The primary aim of the treatment plan is the resolution of SRF and the consequent improvement of VA, whereas a simultaneous decrease in tumour size is an additional gain. In sight-threatening cases, treatment should be initiated as soon as possible for better visual recovery [1, 7]. In symptomatic cases, especially in sub-foveal CCH or juxta-papillary location, where treatment options like TTT and laser photocoagulation may cause retinal damage, PDT is the treatment of choice. PDT causes SRF reabsorption with the fewest complications and recurrences. Compared to alternative therapies, PDT definitely poses a higher out-of-pocket cost.

Intravitreal injections of anti-VEGF may be less effective in treating CCH than retinal neovascularisations. Still, they offer a higher advantage in being minimally invasive and causing no effects on the surrounding healthy retinal tissue. While there are elevated levels of anti-VEGF factors in retinal neovascularisations, the haemangiomas originate from vascular turbulences; therefore, the suppression by anti-VEGF does not cause tumour regression as in neovascularisation. Thus, the combination therapy of anti-VEGFs with other treatments gives better outcomes in CCH. β-adrenergic receptors are present on capillary endothelial cells and regulate vasodilatation and angiogenic factor pathways. Propranolol is a nonselective β-adrenergic receptor blocker. Therefore, oral propranolol can cause endothelium vasoconstriction, decreased basic fibroblast growth factor (b-FGF) expression, hypoxia-inducible factor 1α (HIF-1α) and VEGF through the RAF-mitogen-activated protein kinase pathway and inhibit endothelial proliferation [8]. As VEGF and b-FGF play an essential role in infantile haemangiomas, propranolol is the first-line treatment in these lesions [9]. Assuming a similar growth pattern between infantile haemangioma and CCH, Sanz-Marco *et al*. [8] described for the first time a case of CCH treatment failure with laser photocoagulation and then successfully managed with oral propranolol. VA improved to 20/20, and the foveal SRF disappeared, resulting in a stable outcome within 8 months. The literature presents mixed results, which remain inconclusive, with some cases of treatment success and others of failure.

Our case highlights the role of managing a peripapillary circumscribed choroidal haemangioma through a minimally invasive approach, combining both treatment options of intravitreal anti-VEGF and oral beta blockers, with successful results.

Conclusion

CCH is a relatively uncommon tumour that is visually impairing owing to its location on the posterior pole and SRF exudation. The aim of treatment in such cases is to improve or preserve the vision. In the absence of large-scale comparative studies, it is difficult to prove the supremacy of any of the treatment options. A minimally invasive, multidisciplinary approach to management proves to be a better strategy in such cases.

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