Case Report

Treatment of Choroidal Hemangioma with Photodynamic Therapy: A Case Report and Literature Review

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Received: July 18, 2018; Accepted: August 27, 2018; Published: September 03, 2018

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Abstract

A 22 year-old-woman complaining of bad vision in the right eye, which only had light perception was presented at our hospital. Fundus examination showed a 2 disc diameter (DD) orange-red elevated lesion temporal to a 6 DD central serous retinal detachment. The patient was diagnosed of circumscribed choroidal hemangioma with serous retinal detachment and was treated with intravitreal anti-vascular growth factor plus photodynamic therapy. It is a special case of circumscribed choroidal hemangioma because is associated to hemangioma on the right foot.

Keywords: Circumscribed choroidal hemangioma, photodynamic therapy, serous retinal detachment

Introduction

Choroidal hemangioma is a benign choroidal vascular tumor caused by congenital vascular malformation, that is present as two subtypes: circumscribed and diffuse. The diffuse form occurs in association with Sturge-Weber syndrome. Circumscribed choroidal hemangioma manifests as an isolated unilateral tumor without systemic associations, induces vision loss when it is located in the macula or it leaks fluid which causes retinal detachment [1-3].

Circumscribed choroidal hemangioma is considered rare, the incidence of the disease is difficult to estimate since most patients come to medical attention when they become symptomatic or the tumor is discovered during ocular examination. The differential diagnosis is of high importance due to the probability of several posterior segment lesions such as central serous chorioretinopathy, posterior scleritis, choroidal melanoma, choroidal metastasis because of the risk of ocular and systemic morbidity and mortality [4]. Since CCH can mimic other ocular conditions, additional testing is required for proper diagnosis, these tests include fluorescein angiography used to locate the tumor and leakage, ultrasonography used to measure the tumor and internal reflectivity and optical coherence tomography (OCT) which is helpful in detecting lesions less than 1mm thick and can be used to evaluate the efficacy of treatment [5,6].

CCH is often asymptomatic, in this case the treatment is observation. Although CCH is benign, it can lead to complications, including macular edema, serous retinal detachment and neovascular glaucoma. The goal of any treatment is resolution of subretinal fluid, the decrease in tumor size and visual stabilization. Several therapeutic approaches have been developed, including xenon arc photocoagulation, argon laser photocoagulation, transpupillary
thermotherapy, different types of radiation therapy, recently, photodynamic therapy, oral propranolol, intravitreal injection of antivascular growth factor (anti-VEGF) and systemic infliximab infusion have been introduced [3,7-13].

Case Report

A 22 years old lady presented to us with complaints of bad vision in her right eye. She had light perception. Anterior segments of both eyes were normal with equal, round and reactive pupils. Intraocular pressures were within normal limits. Fundus examination of the right eye showed a 2 DD orange-red elevated lesion temporal to a 6 DD central serous retinal detachment with overlying retinal pigmentation and fibrosis. Ultrasonography revealed a 12.5 mm in diameter x 6 mm in thickness choroidal mass on B-scan with high internal reflectivity on A-scan. Fluorescein angiography was performed, it showed hyperfluorescence in all stages. The patient was diagnosed of Circumscribed choroidal hemangioma (CCH) with serous retinal detachment (Figure 1). The lesion was treated with intravitreal antivascular growth factor 1.25mg/0.05ml and was repeated one month later without success. Photodynamic therapy was used with administration of intravenous verteporfin at a dose de 15mg followed by treatment with laser, two spots at 4,000 µ, 83 s and 100 mJ each. retinal fluid resorption and flattening of the tumor was obtained but visual acuity not improved. This condition has sustained two year of follow up (Figure 2). Her medical examination showed hemangioma in the right foot (Figure 1).

Figure 1: (a) Circumscribed choroidal hemangioma with serous retinal detachment, (b) fluorescein angiography showing staining of CCH, (c) ultrasonography showing the size of the tumor and the retinal detachment, (d) hemangioma in the foot
Review of Treatment

In case of asymptomatic choroidal hemangioma without subretinal fluid, observation is indicated. Treatment is necessary when the vision decreased due to secondary exudative retinal detachment, macular edema and subfoveal fluid [14-16].

The traditional treatment was xenon arc photocoagulation, it produced resolution of subretinal fluid but left scarring that limit visual recovery. After the argon laser became preferred, Shields et al. reported 62% resolution of subretinal fluid and 71% stability of vision [17] argon laser also cause scarring and retreatment are often necessary because subretinal fluid tends to recur [1].

Another treatment is transpupillary thermotherapy (TTT) utilizes 810nm diode laser with a large spot size and long exposure time causing hyperthermia and irreversible cytotoxic effect, sclerosis of vascular channels, this leads to a partial or complete tumor regression, the use is limited to extrafoveal post-equatorial CCH less than 10mm in diameter and 4mm in thickness [18]. Cennamo et al. evaluated the effectiveness and safety of TTT in 25 eyes with CCH, all tumors regressed after treatment, visual acuity was stable in 85% and tumor thickness decreased from 3.52 to 1.8 mm, 85% of patients were treated once, 10% twice and 5% three times. TTT for they is effective and safe for symptomatic CCH not involving the fovea [19]. Wang et al. used TTT to treat 114 eyes with CCH. In their cases visual acuity maintenance with fluid absorption was noted in 66.7% after the treatment [20].

Radiation therapy has been recommended for choroidal hemangiomas with extensive subretinal exudation and retinal detachment where photoagulation cannot be used. The types of radiation therapy include external beam irradiation, episcleral plaque radiotherapy (brachytherapy) and proton beam irradiation. Due to the damage, complications and side effects, these treatments are now eliminated. Naseripour et al. treated 21 eyes diagnosed with symptomatic CCH underwent Ruthenium-106 plaque radiotherapy, from the initial to 1-year follow up, vision improved in 12 eyes, was stable in 7 eyes, and became worst in 2 eyes; subretinal fluid and cystoid macular edema resolved in all eyes, also tumor thickness decreased. Side effects included, radiation-related retinopathy in 5 eyes, radiation-related papillopathy in 1 eye, and subretinal fibrosis in 2 eyes [6]. Zeisberg et al. reported results of 50 eyes with CCH treated with proton beam irradiation, a total dose of 20 Cobalt Gray equivalent was administered, and nine eyes were pretreated by PDT. The mean follow-up was 4 years, tumor thickness decreased in all patients and retinal re-attachment was achieved; 23 eyes developed radiation retinopathy, further complications included sicca syndrome in 9 eyes, cataract in 10 eyes, and radiation optic neuropathy in 4 eyes [10]. Mahdjoubi et al. treated 43 patients with symptomatic CCH by hyperfractionated proton beam therapy, 20 gray relative biological effectiveness in 8 fractions;
retina was reattached in all patients, visual acuity was improved or stabilized in 37 patients, all patients presented regression of tumor thickness [21].

With the introduction of photodynamic therapy (PDT) into the field of ophthalmology, it became the election treatment for CCH. PDT is a laser treatment that, through a photosensitizer like verteporfin is injected into the bloodstream (6mg/m² dose), verteporfin accumulates in the blood vessels in the retina and choroids, is activated by laser energy with a radiant exposure of 100 J/cm², at an irradiance of 600 mW/cm², over an interval of 83 s., allows vascular occlusion and cellular destruction with minimal damage to adjacent structures by triggering the release of free radicals in the areas needing treatment [1,2,22]. The variations to the PDT were the number of spots, single spot or multiple spots, overlapping or not [7,23], the increase of laser power settings [8,24,25], the increase of the duration of exposure [26], double dose of the verteporfin [27]. Published data demonstrated that PDT is safe and effective therapy for CCH (Table 1). Tian et al. compared the effect of indocyanine Green (ICG)-enhanced laser thermocoagulation and PDT in 36 patients with CCH, the results indicated that both methods can lead to tumor atrophy and the absorption of exudation. However, the damage of ICG-enhanced laser thermocoagulation is smaller than that of PDT [28]. Reported complications are membrane epiretinal, choroidal neovascularization and vascular occlusion [29,30].

Table 1: Review of treatment with PDT.

<table>
<thead>
<tr>
<th>No. Of Patients</th>
<th>Treatment</th>
<th>Results</th>
<th>Reference</th>
</tr>
</thead>
<tbody>
<tr>
<td>25</td>
<td>PDT 6mg/m² 689 nm 100J/cm² (22) 50J/cm² (3)</td>
<td>Three eyes treated with 50J/cm² received a second PDT session at 100J/cm²</td>
<td>[4]</td>
</tr>
<tr>
<td>16</td>
<td>PDT 1.69 sessions</td>
<td>Three eyes needed TTT, intravitreal ranibizumab, dexamethasone intravitreal implant</td>
<td>[14]</td>
</tr>
<tr>
<td>13</td>
<td>PDT 1 session (7)</td>
<td>2 sessions (5), 5 sessions (1) and was finally treated with EBRT</td>
<td>[15]</td>
</tr>
<tr>
<td>25</td>
<td>PDT 50J/cm² (18 subfoveal) 75J/cm² (7 perifoveal)</td>
<td>Two subfoveal were re-treated</td>
<td>[16]</td>
</tr>
<tr>
<td>22</td>
<td>PDT Overlapping spots at 50J/cm² for 83s (14) Single spot at 5J/cm² for 166s (8)</td>
<td>No significant differences in tumor regression were found between the two groups</td>
<td>[23]</td>
</tr>
<tr>
<td>20</td>
<td>PDT Standard 6mg/m² verteporfin infusion in 10min, treatment at 15 min; 50J/cm² for 83s Bolus 6mg/m² verteporfin infusion bolus in 1 min; treatment at 5 min; 100J/cm² for 166s</td>
<td>Both bolus and standard PDT induce tumor regression</td>
<td>[24]</td>
</tr>
<tr>
<td>4</td>
<td>PDT</td>
<td>Three re-treatments (2). Four re-treatments (2)</td>
<td>[25]</td>
</tr>
<tr>
<td>27</td>
<td>PDT with double duration</td>
<td>Stopped exudation (70%) and foveal thickness decreased</td>
<td>[26]</td>
</tr>
<tr>
<td>17</td>
<td>PDT with double dose (10) and standard dose (7)</td>
<td>Double dose provided better tumor regression</td>
<td>[27]</td>
</tr>
</tbody>
</table>

PDT: Photodynamic Therapy; TTT: Transpupillary Thermotherapy; EBRT: External Beam Irradiation Therapy
Intravitreal injection of anti-VEGF medication (bevacizumab 1.25 mg/0.05ml) has been used to treat edema macular secondary to CCH, is not invasive and has not effect on the surrounding retinal tissue, one drawback is that it needs to be repeated often. Kwon et al. treated 12 patients with CCH; 8 patients with TTT as primary treatment, 6 showed complete resolution of serous macular detachment, 1 had no recurrence and 5 had recurrence 32.8 months later. The duration of treatment with bevacizumab appears to be short [9]. Mandal et al. treated 3 patients with CCH by intravitreal bevacizumab, all the patients had complete resolution of the serous retinal detachment and was maintained at least 12 months. Intravitreal bevacizumab may be used in combination with TTT or PDT, this combination treatment is safe and more effective [11,12,31].

Oral propranolol therapy was used for patients with CCH at 10 mg 3 times a day and was increased monthly until the desired effects were observed. Sanz-Marco et al. reported a case of CCH treated with oral propranolol, whereupon visual acuity improved, and the macular detachment resolved [13]. Tanabe et al. used oral propranolol hydrochloride to treat 5 patients with CCH. Although this therapy may improve the condition of patients, it does not seem to have a critical therapeutic effect [32]. Sancho et al. administered oral propranolol at a dosage of 1.5mg/kg/day to 5 patients with CCH, the study showed that propranolol is not effective as monotherapy in the treatment of CCH [33].

**Conclusion**

The patient was safely treated with anti-VEGF plus PDT. Although this patient had advanced visual deficit and minimal visual potential was treated because we considered that subretinal fluid may lead to neovascular glaucoma and the need for enucleating. This is a special case where the CCH is associated to hemangioma in the foot. The management of CCH should be tailored to the individual patients including the tumor location, size and presence of serous detachment. The treatment goal is resolution of subretinal and intraretinal exudation, also tumor regression or resolution. PDT has showed be the best therapy for CCH.

**References**


