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# Periocular Infantile Haemangioma and the Role of Propranolol

Tanveer Anjum Chaudhry, Mustafa Kamal and Khabir Ahmad

## ABSTRACT

This case of a four and a half-month-old girl describes periocular infantile haemangioma which was treated successfully with propranolol. She developed a red coloured lesion around the left upper eye lid at one week of age. A gradual increase was noticed in the size of the lesion and by the age of four and a half months, the swelling had increased enough to cover her visual axis, completely occluding her left eye. Oral propranolol therapy was initiated with a daily dosage of 1 mg/kg body weight. The dose was increased gradually, and there was an excellent response to propranolol treatment. A complete eye opening was observed 8 months after the initiation of this treatment.

Key Words: Periocular haemangioma. Infantile. Propranolol. Efficacy.

### **INTRODUCTION**

Capillary haemangiomas are the most common childhood eye tumours (periocular and orbital) that typically appear in infancy but most tend to decrease in size overtime.1 Periocular haemangiomas are 3 times more common among females than males, and can be familial. They can cause refractive and occlusive amblyopia if not treated timely. Most of these tumours regress completely without any residual evidence in 3 to 7 years time. The reddish lesion slowly changes to gray colour and the surface epithelium slowly changes to a more normal skin appearance with thin and wrinkled texture. These changes are associated with skin hypotrophy. Oral steroids and intra-lesional steroid injections are the most common treatment options for such haemangioma.<sup>2</sup> However, there is no standard treatment for these lesions. Propranolol is a new and effective addition to its treatment.3-5

It is postulated that basic fibroblast growth factor, also known as bFGF and vascular endothelial growth factor (VEGF) are responsible for the development and persistence of infantile haemangiomas. Propranolol produces initial vasodilatation and relieves ischaemia of the affected area. Therefore, it reduces the expression of genes responsible i.e. bFGF and VEGF by down regulating Raf-mitogen-activated protein kinase pathway resulting in constriction of pre-capillary sphincters, thus blocking blood flow to the capillaries and gradual resolution of the lesion.

Below is the first report of propranolol use for the treatment of periocular haemangiomas in our country.

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### **CASE REPORT**

A two and a half-month-old girl was brought to the eye clinic of the Aga Khan University Hospital, Karachi with a history of having developed a red-coloured lesion around the left upper eyelid at one week of age. There was a large left periocular swelling covering the entire upper eyelid. However, she was able to open her left eye enough to reveal her pupil and the red reflex (Figure 1). The swelling was purple in colour, with smooth compressible surface. Valsalva maneuver resulted in enlargement of the swelling. The other eye and its periocular area were normal. The baby was following light and fixating on objects, with normally reacting pupils. She was kept under observation, but by the age of four and a half months, the swelling had increased enough to cover her visual axis and completely occlude her left eye.

Before starting of treatment, an informed consent was obtained from the parents of the patient. Her pretreatment physical examination, laboratory work-up, chest X-ray and ECG were normal. Magnetic resonance imaging (MRI) brain and orbits were advised but were refused by the parents.

She was hospitalized under the care of a paediatrician and oral propranolol therapy initiated with a daily dosage of 1 mg/kg body weight. The dose was slowly increased to 4.5 mg/kg body weight/day (in three divided doses) over the next few days. Her blood pressure and respiratory rates were monitored. Her blood sugar was also measured daily to monitor propranolol-related hypoglycaemia. While there was very little response to this therapy in the first two weeks, there was a marked response at 1 month. Prednisolone with dose of 2 mg/kg body weight/day was added for a short period (6 weeks) to accentuate reduction in the size of haemangioma but was gradually reduced and eventually stopped due to its side effects such as weight gain, puffiness, gastric upset, skin eruptions on back and thighs, polyphagia, polydipsia and polyuria. She was examined on a



propranolol.



**Figure 2:** Three and a half months after the initiation of treatment with propranolol.

monthly basis. Two months after the initiation of treatment, she could open her affected eye only slightly. Three and a half months later, she was able to open her eye well enough to reveal more than half of the cornea (Figure 2). At 8 months, the response to therapy was excellent with complete eye opening and only a small remnant of haemangioma (Figure 3). There were no significant propranolol-related adverse events except one episode of difficulty in breathing which resolved within couple of days.

#### DISCUSSION

Currently, there is no standard treatment for haemangiomas. In 2008, Leaute-Labreze first described the role of propranolol for the treatment of severe haemangiomas of infancy.<sup>6</sup> Since then, there has been a great interest in the use of propranolol for the treatment for infantile haemangiomas. Propranolol alone or in combination with existing treatment modalities such as steroids is being explored as a new and effective treatment option.

We treated the child with propranolol and response to the therapy was excellent. An almost complete reduction in size of haemangiomas was achieved after 8 months of treatment with only a small, flat remnant of it left. There was complete eye opening. The standard dose of propranolol for the treatment of infantile periocular haemangiomas has not been defined to-date. Previous investigators have reported successful outcomes with dozes ranging from 1 mg/kg body weight to 5mg/kg body weight.<sup>7</sup> This child was treated with propranolol therapy beginning at a dose of 1 mg/kg body weight/day and slowly increasing to 4.5 mg/kg body weight/day over the next few days. The child was regularly monitored by a paediatrician for any adverse event. Only one episode of difficulty in breathing was observed which resolved within couple of days. We recommend that drug should only be given when its benefits outweigh its known risks.

fter the Figure 3: Eight months after the initiation of treatment with propranolol.

A limitation of this case report is that it was unclear if this haemangiomas involved the orbit. MRI orbits were advised but were refused by the parents.

To the best of authors' knowledge, this is the first reported use of propranolol in the treatment of periocular haemangiomas in Pakistan. This case report as well as many other illustrate the advantages of this medicine in the treatment of periocular haemangiomas. The therapeutic use of propranolol is important as it minimizes the need for systemic steroid, resulting in minimum side effects. Moreover, its use is likely to reduce the need for any surgical intervention.

Periocular haemangiomas are relatively uncommon conditions. Although self-limiting in many cases, it may leave permanent blemish which may be associated with stress and anxiety for the parents. Effective treatment is important to minimize the potential for blemish in cosmetically important anatomic sites, and to minimize the risk of functional visual deficit.

At present, there has been no consensus regarding what should be the dose and duration of propranolol treatment for this condition. Large randomized trials are needed to determine whether treating periocular haemangiomas with propranolol is effective.

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