CASE REPORT

Treatment of complex infantile haemangioma in a resource-poor setting

Ronald Natawidjaja,¹ Ewen Wang²¹Alam Sehat Lestari Clinic, West Kalimantan, Borneo, Indonesia²Division of Emergency Medicine, Stanford University School of Medicine, Stanford, California, USA

Correspondence to

Dr Ewen Wang,
ewen@stanford.edu

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SUMMARY

Infantile haemangiomas affect approximately 5% of the population and usually do not require treatment. However, complex cutaneous haemangiomas can cause disabling disfigurement, while haemangiomas in the brain, airway or gastrointestinal tract can cause life-threatening complications. Although children with infantile haemangiomas are often first brought to general practitioners and paediatricians by parents for care, they are thought of as a surgical problem and usually referred to specialty care. We present a case of an infant from a resource-poor setting in rural Indonesia with disfiguring facial haemangiomas, as well as a probable airway haemangioma causing stridor at rest. The infant was treated with oral propranolol with marked involution of the cutaneous haemangioma, resolution of stridor and increase in weight.

BACKGROUND

Infantile haemangiomas are the most common tumours of infancy and are not usually clinically important. However, in some cases, cutaneous haemangiomas can cause permanent disfigurement; haemangiomas in the brain, airway and gastrointestinal tract can cause life-threatening complications. General practitioners and paediatricians generally are not taught how to treat infantile haemangioma, and cases are usually referred to specialists such as otolaryngologists and dermatologists.

We present the case of an infant with complicated infantile haemangiomas in a resource-poor setting. The infant had no option for subspecialty care. After a review of the literature and phone consultation with a paediatric otolaryngologist, she was treated with oral propranolol, her only option for treatment. After only 1 month of treatment, the haemangioma had shrunk, the stridor had resolved and the child had gained 1.4 kg. After 6 months of treatment, the haemangioma had flattened and shrunk markedly with minimal disfigurement. Treatment was stopped with no regrowth of the haemangioma.

Oral propranolol is becoming a first-line treatment for complicated infantile haemangiomas even when surgery is available. In resource-poor settings, propranolol may be the only treatment option, with life-changing/saving outcomes. It is important for general practitioners who care for children in these settings to be aware of this non-surgical treatment option for complicated infantile haemangiomas.

CASE PRESENTATION

An 11-month-old girl was brought by her mother to a rural clinic in Borneo, Indonesia. The child

had stridor at rest, as well as multiple facial lesions. The lesions were not present at birth, but had developed from small red spots noted by the mother around 2 months of age. Since then, the lesions had grown in size, and the mother stated that the child had started snoring. At the time of presentation, in addition to the disfiguring facial lesions, the child also had respiratory distress and decreased oral intake (most probably from increased work of breathing). The child had been treated with oral steroids, antibiotics and inhaled β -agonists with no improvement. She was referred to the Indonesian capital city, Jakarta for surgery, but the family had no means to travel and came to our clinic as a last resort.

On physical examination, the patient was small (5.6 kg), alert and active, with stridor at rest but normal respiratory rate and oxygen saturation. The child had multiple bright red, disfiguring lesions on her lower lip, over her left eyelid and right temporal area (figure 1A, B). She had no other lesions noted, and the rest of her physical examination was within normal limits.

DIFFERENTIAL DIAGNOSIS

Other vascular malformations which can present in infants include: congenital haemangiomas, port wine stains and other arterial and venous malformations. However, the diagnosis of infantile haemangioma can usually be based on clinical examination and history. Complex haemangiomas can occur with other structural disease, in particular neurocutaneous syndromes such as: PHACE (posterior fossa abnormalities, haemangioma, arterial malformations, coarctation and other cardiac lesions and eye abnormalities).

TREATMENT

After a review of the literature and consultation with an otolaryngologist by phone, it was decided to start the child on the only treatment available: oral propranolol. Her vital signs were recorded. Full examination revealed no other abnormalities on physical examination and a screening ECG demonstrated normal sinus rhythm with no abnormalities. The infant was started on oral propranolol 1 mg/kg/day divided into three doses (10 mg tablet was crushed and reconstituted). The patient was observed for 3 h after the first dose. Parents were educated to look for signs of complications of the treatment (hypoglycaemia, bradycardia and hypotension manifested clinically as lethargy and poor perfusion). They were also

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Figure 1 The patient prior to treatment (A) front profile and (B) left profile.

Figure 2 The patient after 1 week of treatment (A) front profile and (B) left profile.

instructed to give the medication with food and to stop the propranolol if the child had poor feeding.

OUTCOME AND FOLLOW-UP

At 1 week follow-up, the patient's stridor had decreased and her appetite had improved. Propranolol dosing was increased to 2 mg/kg/day (figure 2A, B) and the child was observed in the clinic for

3 h after the first increased dose. The patient was followed in the clinic weekly. After 6 weeks, the child's stridor had resolved and her weight had increased to 7 kg (figure 3A, B). The patient was then followed up every 3 months with continued weight gain, involution and fading of the facial haemangiomas. Six months after treatment, her propranolol dosing has been tapered off with no change in size of the haemangiomas (figure 4A, B).

Figure 3 The patient after 6 weeks of treatment (A) front profile and (B) left profile.

Figure 4 The patient after 6 months of treatment (A) front profile and (B) left profile.

DISCUSSION

Infantile haemangiomas have a prevalence of approximately 5–10%.¹ Most infantile haemangiomas are benign, solitary, self-limited and do not require treatment.² Infantile haemangiomas appear approximately 2–4 weeks after birth.³ Although they can occur anywhere, the majority are found on the head and neck. The typical haemangioma grows rapidly during the proliferation phase (first 3–6 months of life), and then regresses during the involution phase (course of years). As the haemangioma involutes, the bright red colour fades and it flattens. However, involution does not result in normal skin. Scarring, atrophy and continued discoloration compared to normal skin are common. Ulceration can occur in large lesions.

Multiple haemangiomas occur in approximately 20% of infants with infantile haemangioma.⁴ Haemangioma characteristics including size and location can predispose infants to complications including permanent disfigurement. Visceral haemangiomas can cause complications depending on the specific organ involved. Complications include high output heart failure, respiratory distress and CNS sequelae.^{5–7} In these cases treatment is indicated to reduce morbidity and prevent or minimise complications.⁸

The effect of propranolol on involution of infantile haemangiomas was discovered incidentally in 2008 when a child with obstructive hypertrophic myocardiopathy being treated with propranolol demonstrated regression of a facial haemangioma.⁹ Since then, propranolol has been introduced as a primary treatment for complicated haemangiomas even by specialists without resource constraints, as surgical excision of these lesions can still result in significant morbidity.^{10–13}

In resource-poor settings, subspecialty consultation and surgical treatment are not often available; however, propranolol is commonly available and safe to use with careful monitoring and parental education. The exact mechanism is not known, although possible mechanisms include: vasoconstriction and decreased expression of endothelial growth factor.¹⁴

Patient's perspective

The parent felt very grateful to be treated in ASRI because they had already been to many medical care centres and had even been to a specialist with no improvement.

Learning points

- ▶ General practitioners caring for children in resource-poor settings should be aware of propranolol as a first-line (and often only) option of treatment for complicated infantile haemangiomas.
- ▶ Propranolol should be started after screening vital signs, and complete examination for stigmata of associated lesions.
- ▶ The patient should be monitored after starting or increasing propranolol for 3–4 h for cardiovascular side effects. Parents should be informed about possible complications of β -blocker therapy and be educated to stop treatment if the child is ill or not feeding well.
- ▶ Propranolol is typically tapered off after 6 months of treatment or at 1 year of age after the haemangiomas appear to have stopped responding. It can be restarted if the haemangiomas start to grow again.

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