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INFANTILE HEMANGIOMA OF PHILTRUM MANAGED CONSERVATIVELY WITH ORAL PROPRANOLOL: A CASE REPORT WITH LITERATURE REVIEW

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Abstract

This case report aims to provide a detailed examination of an infantile haemangioma case, shedding light on its clinical presentation, diagnostic approach, and therapeutic considerations. By delving into the intricate nuances of this condition, we hope to contribute valuable insights into the evolving landscape of paediatric dermatology and enhance our understanding of the multifaceted nature of infantile haemangiomas.

INTRODUCTION

Infantile haemangiomas, the most common benign vascular tumours of infancy, present a unique and intriguing challenge in the realm of paediatric dermatology. These proliferative vascular lesions, often appearing shortly after birth, exhibit a characteristic pattern of rapid growth followed by spontaneous involution, making their clinical course both dynamic and variable. While most infantile haemangiomas pose minimal medical concern, a subset may necessitate intervention due to complications or cosmetic considerations.

They are more common in females, premature infants, and those with low birth weight. The incidence varies, but it is estimated to affect up to 4-5% of infants.{1}

The exact aetiology of infantile haemangioma remains unclear, but recent research has suggested a multifactorial process involving genetic, hormonal, and environmental factors. The role of endothelial progenitor cells and the renin-angiotensin system is also under investigation. {2}

Infantile haemangiomas exhibit a characteristic clinical course, with rapid growth during the first few months of life followed by a spontaneous involution phase. However, some

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lesions may cause complications such as ulceration, obstruction of vital structures, or aesthetic concerns. {3}

Various treatment modalities exist, including systemic or topical beta-blockers (propranolol), laser therapy, and surgical excision. The choice of treatment depends on the size, location, and complications associated with the haemangioma. {4}

Complications associated with infantile haemangiomas include ulceration, airway obstruction, congestive heart failure (in large, hepatic haemangiomas), and potential psychological impact due to disfigurement. {5}

Most infantile haemangiomas resolve without sequelae, but close monitoring is necessary for those with complications or high-risk features. Long-term follow-up studies are essential to understand the impact of these lesions on quality of life. {6}

Ultimately, this case report serves as a testament to the ongoing evolution of our understanding and management of infantile haemangiomas. By sharing our experiences and insights, we aspire to contribute to the collective knowledge base, fostering a collaborative environment in which clinicians can continually refine their approach to these enigmatic vascular lesions in the youngest members of our patient population.

Case report

A 9-month-old girl was brought by her parents to our dermatology OPD in a tertiary care hospital in Greater Noida, Uttar Pradesh, India. The child had a single red coloured raised lesion on her upper lip area crossing the midline. The lesion was not present at birth but had developed from small sized reddish raised lesions which the mother had observed at about 2 months of age. The lesions had a history of gradual progression to the size at presentation. The lesion was not associated with any respiratory distress or reduction in oral intake. She had not received any treatment prior to her visit to our OPD.

On physical examination, the patient weighed 8.5 kg and was 70 cm long; which was normal for her age and sex according to WHO growth chart. She had normal respiratory rate, blood pressure and oxygen saturation. She had a solitary bright red vascular nodule on her philtrum in the midline with extension on the left side measuring 2.5 cm*1cm in size. The lesion extended superiorly to the left side of the tip of the nose covering the medial one third of the left nostril. The swelling was firm, did not bleed on touch and was non-tender. Diascopy revealed non-blanchable erythema. On dermoscopic examination, lacunae and red dots were seen during the first visit. Patients clinical presentation and clinic-dermoscopic examination was consistent with the diagnosis of Infantile Hemagioma.

She had no other lesions on her body and rest of the physical examination was within normal limits.

Given the fact that the philtrum is a part of the dangerous area of the face; its infection could have led to spread to the cavernous venous sinus. Since the lesion extended to the tip of the nose, it could have led to breathing difficulty if it increased in size. Lastly,

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cosmetic disfigurement is also undesirable over face. For these reasons we took a decision to start the patient on systemic medication. Since a decade or so, oral propranolol has established itself as a preferred systemic agent for treatment of infantile haemangiomas

Once the decision to start the patient on oral propranolol was taken, her vitals were recorded and paediatric consultation was done to rule out any cardiac abnormalities. Screening ECG revealed normal sinus rhythm with no abnormalities. The patient was started on oral propranolol at a dose of 1mg/kg/day in 3 divided doses. The oral preparation was made by crushing 10 mg propranolol tablets and reconstituted with distilled water. The patient was admitted for 2 days for strict vital monitoring by the team of doctors and to manage any complications that might occur after starting the treatment. After the discharge parents were instructed to continue the medication at home and come for regular follow up. They were counselled about recognizing potential treatment complications, such as hypoglycaemia, bradycardia, and hypotension, which may present as lethargy and reduced perfusion. They were also advised to administer the medication with food and discontinue propranolol if the child experienced feeding difficulties.

The patient was breastfed with supplementary foods being fed by the parents. The mother was advised to breastfeed the baby before propranolol administration to reduce the chances of hypoglycaemia.

Outcome and follow-up

At 1 week follow-up, the patient was tolerating her treatment quite well so the propranolol dosing was increased to 2 mg/kg/day 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 infantile haemangioma had reduced in size considerably. Dermoscopic features also improved with the treatment, the earlier observed lacunae became empty and the erythema got replaced by brown diffuse pigmentation. The patient was then followed up every 3 months with continued weight gain, involution and fading of the facial haemangioma. Six months after treatment, the infantile haemangioma has reduced greatly and the oral propranolol is being tapered off gradually. Due to the midline location of this hemangioma, it is likely to leave behind some residual disfigurement despite appropriate control of the lesion. The patient is expected to benefit from plastic surgery intervention for the same, if desired.

DISCUSSION

Infantile haemangiomas, though often benign, present a spectrum of clinical challenges, emphasizing the importance of a comprehensive understanding for effective management. Our case aligns with existing literature highlighting the variability in presentations, emphasizing the need for individualized treatment approaches. While some infantile haemangiomas may resolve without intervention, others may require therapeutic measures due to complications such as ulceration, functional impairment, or

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disfigurement. In our case, the decision to initiate treatment was guided by the presence of functional impairment, emphasizing the importance of a tailored approach based on clinical presentation.

The evolving picture of therapeutic options for infantile haemangiomas has seen the emergence of non-invasive modalities, such as beta-blockers, which have shown promising results in promoting regression. Our case supports the efficacy of this approach, as the administration of propranolol led to a notable reduction in the size and vascularization of the lesion. This finding aligns with current trends in paediatric dermatology, where beta-blockers have become a mainstay in the management of complicated infantile haemangiomas.

Topical timolol maleate 0.5% gel applied twice a day has also shown to produce a significant reduction in the size of haemangioma in 24 weeks and several studies show a comparative efficacy of topical timolol and oral propranolol. More data is available regarding oral propranolol efficacy in the treatment of infantile haemangiomas, which is why it was our choice of treatment for this patient.

Infants may also have vascular issues like congenital haemangiomas, port wine stains, and various arterial and venous malformations. Yet, diagnosing infantile haemangioma typically relies on clinical examination and medical history. Complex cases of infantile haemangiomas on the face may involve other conditions, notably neurocutaneous syndromes like PHACES (posterior fossa abnormalities, haemangioma, arterial malformations, coarctation, cardiac lesions, eye abnormalities and sternal abnormalities).

REVIEW OF LITERATURE

In the 2012 study by Aletaha M et al[7], 3 infants of 2-3 months of age with infantile haemangioma were treated with oral propranolol solution (Inderal, 20mg/5ml) 2-3 mg/kg per day divided in 2 doses. It was continued till the end of the first year of life and tapered over a period of 2-3 weeks. All infants were followed for 20 months. Lesion size and evolution were assessed during the follow-up period. The result was impressive as significant improvement was noted in all the patients in the first 2 months of treatment with notable results throughout the follow-up period.

In the study done in 2020 by Puri N[8] et al 20 infants with infantile haemangioma were treated with 0.5% timolol and the response was seen when the three drops was applied twice daily over the haemangiomas. The Response to timolol was excellent in 60% of the infants, very good in 20% of the infants, good in 15% of the infants, average in 5% of the infants, and none of the infants showed poor response.

In the study by Boon *et al*., [9] in the study of complications of systemic corticosteroid therapy for haemangiomas, it was found that 44 of 62 children presented with cushingoid facies which began 1 to 2 months after starting at a dose of 2-3 mg/kg/day. It was also observed that children who received therapy for more than 6 months had higher side

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effects; although, the cushingoid facies resolved spontaneously during the final few months of tapering steroids.

In the study by Ng et al. {10} in the study of Laser treatment in infantile haemangiomas, the most common side effect seen in Pulse Dye Laser treatment was seen to be erythema, oedema and purpura. However, these side effects resolved in 1 week in short-pulsed PDL and 2-3 days in long pulsed duration group.

The American Academy of Paediatrics (AAP) also strongly recommends the use of oral propranolol as the first line agent for the medical management of infantile haemangiomas that require systemic treatment. The AAP recommends topical timolol application for thin and superficially located haemangiomas with moderate level of evidence available for the same. {11}

CONCLUSION

The presented case underscores the complex and dynamic nature of infantile haemangiomas, highlighting the importance of individualized management strategies tailored to each patient's unique clinical presentation and needs. Through meticulous examination and consideration of potential complications, such as functional impairment and aesthetic concerns, clinicians can effectively guide treatment decisions, as demonstrated in this case where initiation of oral propranolol led to significant regression of the lesion with minimal side effects. This case adds to the growing body of evidence supporting the efficacy of beta-blockers in the management of infantile haemangiomas, offering a non-invasive and promising therapeutic option. Furthermore, ongoing research and collaboration within the field continue to refine our understanding and approach to these enigmatic vascular lesions, paving the way for improved outcomes and quality of life for affected individuals.

Images

Picture 1: The patient at the time of initial presentation with infantile hemagioma on left upper lip area, crossing the midline

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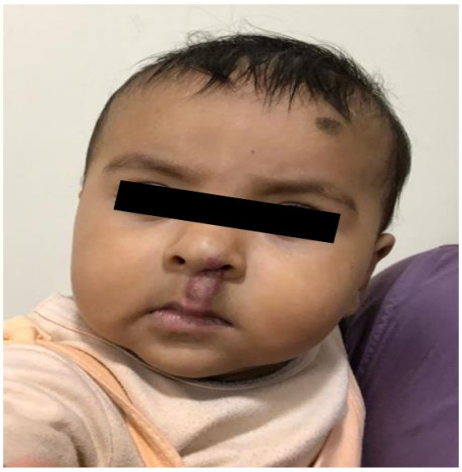
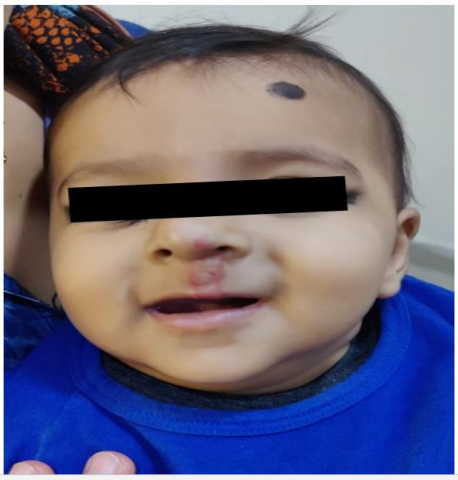


Picture 2: Partial involution of the infantile hemangioma after 3 months of oral propranolol therapy



Picture 3: Significant improvement as seen after 6 months of oral propranolol treatment.

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