

Pleuri-Segmental Ipsilateral Nonsyndromic Infantile Hemangioma Responding Favorably to Oral Propranolol

Sir,

Infantile hemangiomas (IH) may manifest as focal, multifocal, segmental, or indeterminate lesions. Segmental hemangiomas have been associated with an 11-fold associated risk of complications when compared with localized forms and are frequently associated with syndromes.^[1]

A 6-month old girl (preterm with a birth weight of 2.5 kg) was brought to the dermatology outpatient with red vascular lesions on the right side of the forehead, buttock, perineum, and lower limb. At birth, parents noticed faint erythema simultaneously at the abovementioned sites, which progressively increased in size after 1 week. The perineal lesion ulcerated 2 weeks prior to presentation. There was no history to suggest a developmental delay, hypotonia, syncopal attacks, or urogenital abnormalities. Examination revealed well-defined grouped, superficial red vascular papules 0.3–0.8 cm in diameter on the right S1 segment of face [Figure 1a]. Similar lesions present on the right lower limb had a deeper component showing a lacing pattern of blue vascular structures [Figure 1b]. Corresponding labia majora and buttock showed similar lesions with superficial ulceration and scarring at places [Figure 1c]. Ophthalmological examination was normal. Radiologic investigations, including abdominal sonography and magnetic resonance imaging (MRI) of the brain, spine, and pelvis were normal, as were electrocardiogram (ECG), and thyroid function tests (TFTs). She was started on oral propranolol 2 mg/kg/day (crushed tablets), advised local care for ulceration, and showed significant improvement in lesions within 1 month of starting therapy and complete involution after 9 months [Figure 2 a-c].

Segmental hemangiomas constitute around 13–18% of hemangiomas.^[1] As with other segmental lesions, mosaicism has been the proposed mechanism for the occurrence of such hemangiomas.^[2] However, others believe that patterns of segmental hemangiomas (especially of the face) are usually neither blaschkoid/dermatomal nor do they correspond to sensory nerve distributions/known embryonic segments.^[3] Large segmental IHs are often associated with underlying malformations, particularly PHACES and PELVIS/LUMBAR syndromes. Large segmental IH in a “beard” distribution may mark a subglottic hemangioma. Large IHs and hepatic hemangiomas may cause hypothyroidism and may be associated with local complications like



Figure 1: (a) Pretreatment image of segmental hemangioma in the right facial S1 (frontotemporal) region. (b) Pretreatment image of lesions on the right lower limb. (c) Pretreatment image of lesions on the right labia majora and perineal region

ulceration, bleeding, orificial obstruction and cosmetic disfigurement as was seen in our patient.

The occurrence of hemangiomas in multiple segments, as seen in the index case is a rare occurrence. Khanna *et al.* reported the occurrence of large segmental hemangiomas



Figure 2: (a) Posttreatment images of segmental hemangioma in the right facial S1 (frontotemporal) region. (b) Posttreatment images of lesions on the right lower limb. (c) Posttreatment images of segmental hemangioma in right labia majora and perineal region (Note the healing with scarring reminiscent of previous ulceration)

of the right upper limb and shoulder as well as contralateral thigh with high-output cardiac failure. Improvement was seen in cutaneous and hemodynamic symptoms after propranolol therapy.^[4] Dakshayini *et al.* reported a case with segmental hemangiomas on the left side of the face, left upper and lower limbs with PHACES syndrome.^[5] Fortunately, no such syndromic

associations or systemic complications were seen in the present case.

Although all segmental hemangiomas *per se* may not always necessitate medical intervention, it is indicated in patients with complicated IH. Propranolol has surpassed oral steroids in becoming the drug of choice in treating IHs. Our patient responded favorably to the same without any serious adverse effects. Concluding, pleuri-segmental hemangiomas are a rare presentation of IH. Although they often raise a strong suspicion of local and/or systemic complications, they may be nonsyndromic and respond favorably to oral propranolol.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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References

1. Leaute-Labreze C, Prey S, Ezzedine K. Infantile haemangioma: Part I. Pathophysiology, epidemiology, clinical features, life cycle and associated structural abnormalities. *J Eur Acad Dermatol Venereol* 2011;25:1245-53.
2. Yu X, Zhang J, Wu Z, Liu M, Chen R, Gu Y, *et al.* LUMBAR syndrome: A case manifesting as cutaneous infantile hemangiomas of the lower extremity, perineum and gluteal region, and a review of published work. *J Dermatol* 2017;44:808-12.
3. Phan TA, Adams S, Wargon O. Segmental haemangiomas of infancy: A review of 14 cases. *Australas J Dermatol* 2006;47:242-7.
4. Khanna D, Chakravarty P, Arora P, Jain R, Mittal M. Large multifocal cutaneous hemangioma along lines of Blaschko

with cardiac failure treated with propranolol. *Pediatr Dermatol* 2015;32:e18-22.

5. Dakshayani B, Benakappa A. Multiple segmental hemangiomas over one half of the body-A rare feature of PHACES syndrome. *Indian J Pediatr* 2016;83:83-4.

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