Case Report

Infantile Hemangioma with Intracranial Vascular Anomalies: A Case Report

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Abstract

Infantile hemangioma (IH) is a benign tumor of the vascular system that most commonly occurs in infants, with approximately 5-10% of infants presenting with the diagnosis. A characteristic feature of IH is that these tumors appear shortly after delivery and proliferate rapidly in the first year of life, followed by gradual involution or spontaneous regression. Most hemangiomas are cutaneous in origin and self-limiting; however, large hemangiomas in the head and neck may be associated with intracranial vascular anomalies and developmental delay. Herein, we report a rare case of a large hemangioma involving the head and neck that was associated with an intracranial vascular anomaly and developmental delay in a young infant who received propranolol therapy and was monitored in the pediatric neurology clinic.

Keywords: Infantile hemangioma, intracranial vascular anomalies, propranolol therapy, PHACE syndrome, vascular tumors.

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Introduction

Infantile hemangiomas (IHs) are the most commonly occurring tumors of the vascular system in infancy and are identified by their rapid proliferation of endothelial cells followed by spontaneous regression. The incidence of IH, based on available information, has been reported to be between 5 and 10% in infants. IH is also seen more frequently in females, premature infants, and low birth weight infants.[1] IHs typically develop shortly after delivery, proliferate rapidly in the first year of life, and involute in most cases by 7 years of age.[2] IHs occur most frequently on the head/neck (approximately 60%), followed by the trunk (approximately 25%), and upper/lower extremities (approximately 15%).[3] Clinically, IHs are often superficial in presentation and appear bright red with irregular borders; further, deeper lesions may present with bluish discoloration due to subcutaneous location. During involution, IHs gradually soften, fade in color, and shrink in size.[4] Most IHs are self-limiting (benign); however, large segmental IHs of the head and neck may be associated with structural anomalies (that is, intracranial vascular anomalies) and syndromic conditions (that is, PHACE syndrome).[5] These associations emphasize the importance of early identification and evaluation. In this report, we describe a rare case of a large IH of the head/neck with developmental delay and associated intracranial vascular abnormalities.

Case Presentation

A young infant was referred to the pediatric clinic due to a progressively enlarging hemangioma over the head and neck region, which had developed shortly after birth and progressively increased in size over the next few months. The lesion was red, extensive, and of vascular nature, as identified by physical examination. In addition to displaying a large IH of the head/neck, the infant also experienced developmental delays for age. Due to the extensive involvement of IH, further evaluation was undertaken to evaluate associated anomalies. Neuroimaging revealed multiple intracranial vascular anomalies, suggesting the possibility of central nervous system involvement. Evaluation of the clinical and neuroimaging findings supported the diagnosis of a large IH of the head/neck associated with intracranial vascular anomalies. Given the findings, we initiated treatment with oral propranolol, as it is now considered the preferred treatment for complex IHs. After starting treatment, the baby experienced a significant reduction in the size of the IH, particularly in the head/neck area. The child tolerated propranolol therapy well and experienced no significant complications from the medication. Currently, the child continues to receive regular follow-up from the pediatric neurology service for monitoring of neurological outcomes and developmental milestones.

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Discussion

IHs are benign vascular tumors that occur due to the abnormal proliferation of endothelial cells. The typical clinical course of IH includes proliferation during infancy, followed by spontaneous involution (in most instances).[6,7] Although most IHs are isolated cutaneous tumors, large segmental IHs that occur in the head and neck may be associated with intracranial vascular anomalies and the presence of additional structural and functional anomalies. MRI is an important diagnostic test used to assess and monitor for the presence of these conditions. The number of infants with IHs who also have intracranial vascular lesions is likely to be underestimated.[8] A systematic review of 436 patients found that 5% of the patients with IH also had an associated intracranial vascular anomaly; the intracranial vascular anomalies included hemangiomas in the brain, structural brain deformities, and vascular malformations of the brain.[7] Intracranial IHs are also very rare. In a recent literature review, only approximately 44 cases of intracranial IH have been reported. The clinical presentation of intracranial IH can vary significantly, depending on the size and location of the tumor, the patient's neurological function, and structural integrity. Neurological deficits resulting from areas of the brain affected by an IH can range from no deficits, to severe deficits, to death.[9] An additional consideration for patients with large IH of the face is the possibility of the presence of PHACE syndrome, which includes multiple components, including brain malformations, IH, vascular malformations (arteries), congenital heart defects, and ocular defects. Neuroaxial IHs have occurred more frequently in patients with these syndromes than in patients without PHACE syndrome, thus indicating the importance of complete imaging evaluations.[6] Because of the ability of propranolol to significantly reduce the size and vascularity of IHs, and because of its relatively few complications compared with the use of other therapies (steroids, laser therapy), propranolol has become the first-line treatment for complex IHs.[10,11] In this particular case, the timely initiation of propranolol therapy resulted in significant clinical improvement of IH and highlighted the need for ongoing neurologic follow-up related to the patient's intracranial vascular anomalies.

Conclusion

Large segmental IHs occurring in the head or neck may be associated with the presence of intracranial vascular lesions and associated syndromic conditions, including PHACE syndrome. Therefore, timely identification of these lesions, initiation of appropriate imaging studies, and initiation of appropriate treatment are critical to preventing further complications. Currently, propranolol is considered safe for the treatment of IH. All children with associated neurological anomalies should receive long-term neurological follow-up.

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Conflicts of interest

There are no conflicts of interest.

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