https://doi.org/10.5021/ad.2021.33.4.390

Deeply Located Infantile Hemangioma with Rapid Growth Resembles Malignancy

Dae-Lyong Ha[1,3,4], Gi-Wook Lee[1,3], Kihyuk Shin[1,2], Hyun-Chang Ko[1,2], Byung-Soo Kim[1,3], Moon-Bum Kim[1,3], Hoon-Soo Kim[1,3]

[1]Department of Dermatology, School of Medicine, Pusan National University, Busan, [2]Department of Dermatology, Pusan National University Yangsan Hospital, Yangsan, [3]Biomedical Research Institute, Pusan National University Hospital, Busan, [4]Department of Dermatology, School of Medicine, Kyungpook National University, Daegu, Korea

Dear Editor:

The unexpected finding of a growing lump in an infant induces parental anxiety and results in an urgent visit to the hospital. There are five clinical danger signs that suggest the possibility of a malignant tumor in children, namely, 1) onset at birth or in the infantile period, 2) a history of rapid and progressive growth, 3) a firm mass >3 cm in diameter, 4) skin ulceration, and 5) fixation to deep tissue or location below the fascia. However, before attempting to apply these criteria, infantile hemangioma (IH) should be excluded[1].

A 6-month-old girl visited the pediatric orthopedic department of Pusan National University Hospital with a huge protruding subcutaneous tumor on the right upper part of the back, measuring approximately 10 cm in diameter (Fig. 1A). The tumor abruptly appeared 2 weeks ago and enlarged. Magnetic resonance imaging (MRI) showed a lobulated hypervascular mass with internal neovascularization in the subfascial layer adjoining the deep fascia. The tumor exhibited iso- to low-signal intensity in T1-weighted imaging (Fig. 1B) but weak high signal intensity in T2-weighted imaging (Fig. 1C). Based on these findings, the possibility of malignancy was considered by the radiologist and orthopedic surgeon, and exploratory surgery was planned. The patient was referred to our department for consultation to exclude other possible conditions before surgery. As the baby looked well and the tumor was soft on palpation and movable, incisional biopsy was performed. The tumor was detected under the fascia of the trapezius muscle. Histopathologic examination showed well-defined masses composed of multilobular proliferation of numerous vessels (Fig. 1D). Immunohistochemical staining revealed that the endothelial cells showed positivity for CD-31, GLUT-1 and negative for D2-40 stain (Fig. 1E).

After the diagnosis of IH was confirmed, the patient received oral propranolol at a dose of 2 mg/kg/day. The tumor volume decreased dramatically within 3 weeks. Follow-up of the patient revealed that the tumor almost completely resolved after 1 year of medication without any complications (Fig. 1F) and no recurrence of tumor was detected after 2 years. We received the patient parent's consent form about publishing all photographic materials.

Imaging tests can help to diagnose tumorous conditions. However, the success rate of MRI at determining whether a soft tissue tumor is benign or malignant varies between 30% and 90%[2]. Deep-type IH was not apparent in the immediate newborn period but became evident a few weeks after birth. The prolonged growth pattern and minimal overlying skin changes pose clinical challenges[1,3]. Although IH is the most common benign vascular tumor of infancy, there have been no reports using the name subfascial IH. However, it is not completely uncommon because there are cases that are thought to have occurred at that depth in the research examining the imaging findings of IH[4]. Although GLUT-1 was partially expressed in this patient, the expression of GLUT-1 itself is specific to IH unlike other vasculr tumors or malformations[1,5].

Findings from this case emphasizes that deep-type IH

Received April 8, 2020, Revised August 11, 2020, Accepted for publication August 17, 2020

Corresponding author: Hoon-Soo Kim, Department of Dermatology, School of Medicine, Pusan National University, 179 Gudeok-ro, Seo-gu, Busan 49241, Korea. Tel: 82-51-240-7338, Fax: 82-51-245-9467, E-mail: suekimm@hanmail.net
ORCID: https://orcid.org/0000-0002-7649-1446

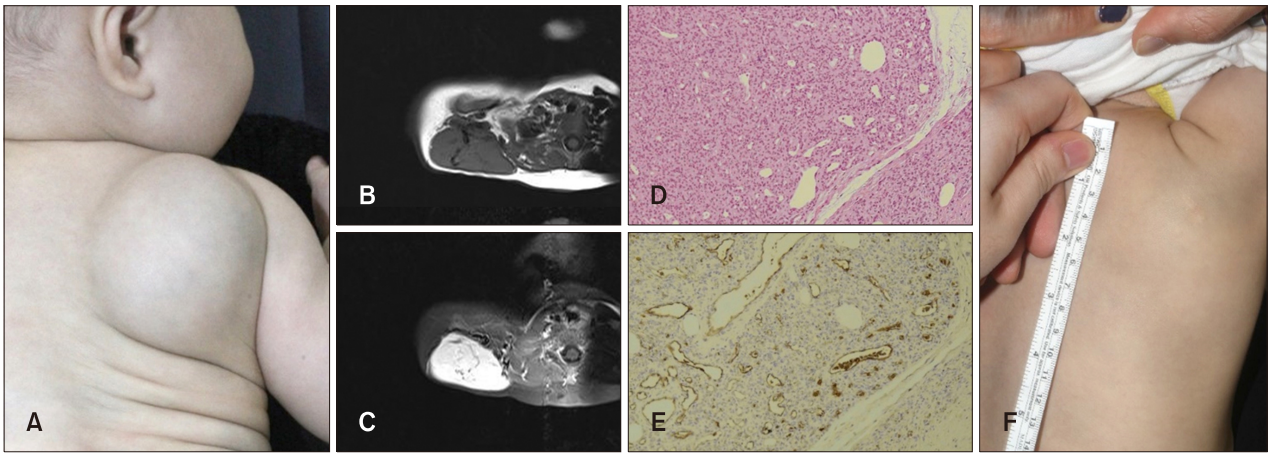

Fig. 1. (A) Protruding skin-colored tumor on the right upper back without skin change. (B) Magnetic resonance imaging showing a lobulated hypervascular mass with internal neovascularization in the subfascial layer adjoining the deep fascia. T1-weighted imaging showing iso to low-signal intensity of the tumor. (C) T2-weighted imaging showing weak high-signal intensity of the tumor. (D) Histopathologic examination shows well-defined non-capsulated masses composed of multilobular proliferation with numerous small vascular spaces lined by plump endothelial cells (H&E, original magnification ×100). (E) Immunohistochemical staining showing GLUT-1 reactivity limited mainly to the luminal border of the endothelial cells in a membranous pattern (GLUT-1 staining, original magnification ×100). (F) After 1 year of treatment with propranolol without any complications.

should be considered when a soft tissue mass displays rapid growth and hypervascularity in infancy.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

FUNDING SOURCE

This work was supported by clinical research grant from Pusan National University Hospital in 2020.

ORCID

Dae-Lyong Ha, https://orcid.org/0000-0002-2268-4795
Gi-Wook Lee, https://orcid.org/0000-0002-5508-8498
Kihyuk Shin, https://orcid.org/0000-0001-8955-9828
Hyun-Chang Ko, https://orcid.org/0000-0002-3459-5474
Byung-Soo Kim, https://orcid.org/0000-0003-0054-8570
Moon-Bum Kim, https://orcid.org/0000-0003-4837-0214
Hoon-Soo Kim, https://orcid.org/0000-0002-7649-1446

REFERENCES

1. Schachner LA, Hansen RC. Pediatric dermatol. 4th ed. Edinburgh: Mosby Elsevier, 2011:1181-1218.
2. Stein-Wexler R. Pediatric soft tissue sarcomas. Semin Ultrasound CT MR 2011;32:470-488.
3. Brandling-Bennett HA, Metry DW, Baselga E, Lucky AW, Adams DM, Cordisco MR, et al. Infantile hemangiomas with unusually prolonged growth phase: a case series. Arch Dermatol 2008;144:1632-1637.
4. Ding A, Gong X, Li J, Xiong P. Role of ultrasound in diagnosis and differential diagnosis of deep infantile hemangioma and venous malformation. J Vasc Surg Venous Lymphat Disord 2019;7:715-723.
5. Huang L, Nakayama H, Klagsbrun M, Mulliken JB, Bischoff J. Glucose transporter 1-positive endothelial cells in infantile hemangioma exhibit features of facultative stem cells. Stem Cells 2015;33:133-145.