



Infantile Haemangioma of Left Second Toe: A Case Report

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Authors' contributions

This work was carried out in collaboration between all authors. Author GTC designed the study, performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript. Authors MFBY and SALN managed the analyses of the study. Author MABMA managed the literature searches. All authors read and approved the final manuscript.

Article Information

DOI: 10.9734/AJORR/2018/43330

Editor(s):

(1) Dr. Ikem, Innocent Chiedu, Professor, Department of Orthopaedic Surgery and Traumatology, Obafemi Awolowo University, Ile-Ife, Nigeria.

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Complete Peer review History: <http://www.sciedomain.org/review-history/26387>

**Received 5th July 2018
Accepted 17th September 2018
Published 25th September 2018**

Case Study

ABSTRACT

Infantile haemangiomas represent the most common type of benign tumours of infancy, with a prevalence of 1-10% worldwide. Even though the disorder is mostly harmless, however, the option of treatment is primarily depending on the progression of this tumour. Some studies reported the disorder in many siblings of the same family, with non-affected parents, pointing out a possible genetic immixture. We present a case of a one month-old boy, suffering with a huge painless swelling of the left second toe. X-ray examination showed no bony lesion. Distal interphalangeal joint amputation of left second toe was also performed. The histopathology test result showed pictures constant as haemangioma. To the best of our knowledge, this case is a first report of infantile haemangioma of the toe mimicking vascular haemangioma.

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Keywords: Infantile haemangioma; vascular malformations haemangioma; second toe.

1. INTRODUCTION

Haemangioma is a common benign vascular tumour, closely resembles with normal vessels and can be found in all organs or extremities of the human body [1]. These tumours develop in the period of infancy, only 20% of them being present at birth, with an increased frequency in the female gender, the white race and low birth weight infants [2]. Infantile haemangioma remains a current challenge for the clinicians regarding the risk factors, prognostic factors, pathogenesis, evolution and treatment [1]. The hypothesis of pathogenesis supports the idea that haemangiomas develop by cellular hyperplasia and by the proliferation of endothelial cells [3]. Substantial research has shown that angiogenic and vasculogenic factors stimulate their proliferation [1] and these markers influence all the evolutionary phases of haemangiomas. Infantile haemangioma of the toe mimicking vascular haemangioma has not been reported yet.

2. CASE REPORT

A 1-month-old boy presented to the department of paediatric with a painless swelling of the left second toe since birth (Fig. 1). As mentioned by his mother the patient had swelling of the left second toe that was huge and increasing in size gradually. Covering skin of the swelling was congested and dark blue appearance was seen and complicated with ulceration. X-ray



Fig. 1. Shows initial presentation of lesion

examination showed no bony lesion in it (Fig. 2). The soft tissue around the lesion was firm in consistency. Preoperative examination yielded no final diagnosis. The lesion was amputated at the level of distal interphalangeal joint (Fig. 3). Macroscopically, the lesion measured 20 mm x 10 mm with a firm surface. Microscopic examination showed that the lesion was composed of solid, cellular lobules consisting of plump endothelial cells lining tiny rounded vascular spaces with inconspicuous luminal and vascular proliferation was reported around sweat glands. The pathological diagnosis of the lesion was haemangioma of the soft tissue (Fig. 4).

3. DISCUSSION

Infantile haemangioma represents as one or more tumours of variable dimensions and aspects, with a predilection to the head and neck area [3]. Other areas, as well as organs, can also be affected. In majority of cases, they are singular, only in 10-25% of cases they form multiple tumours [4]. Superficial haemangiomas present as round or oval tumours, lobulated or with a fine surface, more often in the head-neck areas [3], with a size range of 1-25 cm. They are the most dominant type, being encountered in 50-60% of cases [4]. Deep haemangiomas presented as soft, bluish masses, some with superficial telangiectasia, and are the rarest type - 15% [4]. Mixed haemangiomas is a combine aspect of the two above mentioned forms.



Fig. 2. Shows no bony involvement of lesion



Fig. 3. Shows the amputated lesion at the level of distal interphalangeal joint and measured about 20mm x 10mm with necrotic patch noted over distal part of lesion

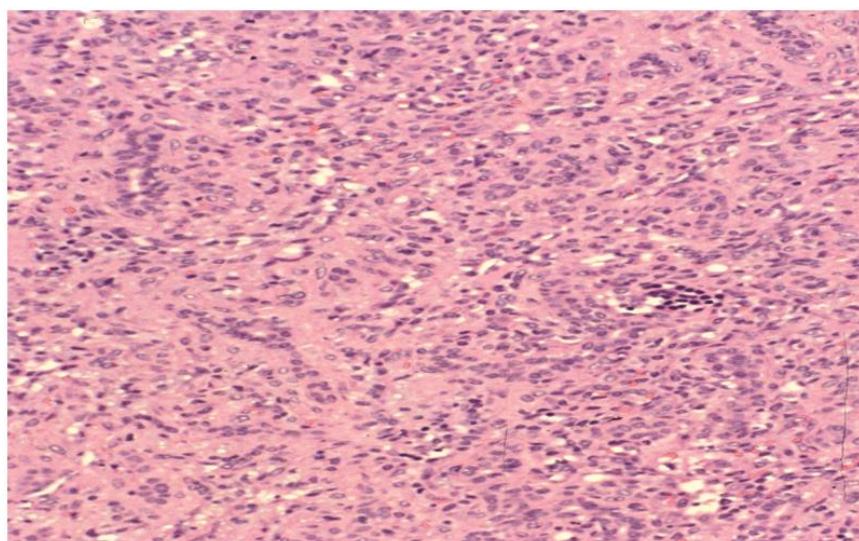


Fig. 4. Shows lesion was composed of solid, cellular lobules consisting of plump endothelial cells lining tiny rounded vascular spaces with inconspicuous luminal and vascular proliferation noted. No atypical endothelial cells are seen

In most of the cases, haemangiomas develop in the first 2-4 weeks of life, but 20% of them can be present during birth. The lesion shows a pale-white to blue-grey macula in the beginning, depending on its profoundness, or a papule with

the same characteristics. They grow during a variable period until 6-9 months, after which they involute, followed by the characteristic evolution. Thus, it has been reported that 50% regression by the age of 5 years and 90% by the age of 9

[1]. The skin and colour will revert to normal condition or remains with a residue of telangiectasia, hypopigmentation, scars or fibrofatty deposits after regression [1].

Approximately 10% of infantile haemangiomas develop complications in their evolution and need individualised therapy [5]. The most common complications are ulceration, infection and haemorrhage. Systemic complications presented with Kassabach Meritt syndrome, characterised by the association of cutaneous and visceral lesions of variable aspects and dimensions, with extracutaneous manifestations, such as consumption coagulopathy [6].

In this case, we need to rule out vascular malformation of haemangioma as it was one of our differential diagnosis because haemangioma is the most frequently encountered vascular soft-tissue abnormality. It has been estimated that haemangiomas comprise 7% of all benign soft-tissue tumours [5]. The most commonly used classification system is mainly based on clinical findings, cellular turnover, and histological features [1]. With this system, vascular anomalies are separated into two major groups: (1) haemangiomas and, (2) vascular malformations [6]. Infantile haemangiomas tend to be small or absent at birth. Shortly after birth, they enter a proliferative phase with a rapid growth that may last for several months, followed by a stationary period and, finally, a period of involution. In contrast, vascular malformations are always present during birth and subsequently enlarge in proportion to growth. They do not involute and remain present throughout the life. Vascular malformations are categorised as lymphatic, capillary, venous, arteriovenous, or mixed malformations on the basis of their histological features [1]. Immunohistochemistry tests for the marker GLUT1 is one of the tool to differentiates the infantile haemangioma from any other type of haemangiomas, or other vascular anomalies but it is not available in our centre.

Preoperatively, we have discussed the usefulness of Magnetic Resonance Imaging (MRI) as one of our investigation tool as it is a useful non-invasive technique for the examination of vascular malformations because of its superior soft-tissue contrast and multiplane capability [6]. MRI plays a significant role in diagnosing, characterising, and determining the extent of the lesions. The MRI appearance of soft-tissue vascular anomalies correlates well

with their biologic classification [2,6]. However, due to robust clinical findings and relevant history from the patient's mother, we assumed that MRI was non-essential for this specific case.

Most of the infantile haemangiomas lend themselves to the "wait-and-see" therapy, but some of them need immediate treatment [5]. Their localisation is one of the essential elements of the therapy approach. Surgery is the indication to disfiguring haemangiomas or multiple-therapy resistant ones [5,7]. If surgery is not indicated early, these disorders become permanent in future [5]. The most important advantage is that it leads to nearly the complete resolution of the haemangioma, but entails risks of bleeding [5]. As for this case, we decided for early distal interphalangeal joint amputation due to the history of progressive increased in size of the haemangioma and intermittent hemoserous discharged from the lesion which is not indicated for conservative management such as the use of propranolol [8] or laser treatment [9].

4. CONCLUSION

Infantile haemangioma of toe may mimic other neoplasm, and could be considered as a differential diagnosis encompasses all factors associated with toe, including osteochondroma, enchondroma, osteoid osteoma, and sarcoma [2]. Therefore, meticulous radiological and clinical investigation is essential to rule out all other possible diagnosis. It should be remembered for the lesions of this region in the human body. To the best of our knowledge, this case report is a first of its kind that describe infantile haemangioma of the toe which mimicking vascular haemangioma.

CONSENT

As per international standard or university standard, patient's written consent has been collected from his guardian and preserved by the authors.

ETHICAL APPROVAL

As per international standard or university standard ethical permission has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

1. Madalina Bota, Gheorghe Popa, Cristina Blag, Alexandru Tataru. Infantile hemangioma: A brief review. *Clujul Med.* 2015;88(1):23–27.
2. Cil Y, Simsek HA, Yıldız H. Primary intraosseous cavernous haemangioma of the toe. *Musculoskeletal Surg.* 2013;97(3): 259-61.
3. Kinoshita G, Matsumoto M, Maruoka T, Shiraki T, Tsunemi K, Futani H, Maruo S. Bone and soft tissue tumours of the foot: Review of 83 cases. *J Orthop Surg* 2002;10:173–178.
4. Lucky AW. Lesiones cutaneas benignas transitorias en el recien nacido. In: Eichenfield LF, Frieden IJ, Esterly NB (Eds). *Dermatología Neonatal*. 2ed. Spain: Elsevier. 2009;85-97.
5. Dahlin DC, Unni KK. Bone tumors: General aspects and data on 8,547 cases. 4th Ed. Charles C. Thomas Pub; Springfield, IL (USA). 1986;522.
6. Joan C. Vilanova, Joaquim Barceló, James G. Smirniotopoulos, Ricard Pérez-Andrés, Miguel Villalón, Josefina Miró, Ferran Martín, Jaume Capellades, Pablo R. Ros. Hemangioma from head to toe: MR Imaging with pathologic correlation. *RadioGraphics*. 2004;24(2).
7. DOI: <https://doi.org/10.1148/rg.242035079>
Bajpai M. Intraosseous vascular tumor of mandible. *J Coll Physicians Surg Pak.* 2016;26(7):638.
8. Hagen R, Ghareeb E, Jalali O, Zinn Z. Infantile hemangiomas: What have we learned from propranolol? *Curr Opin Pediatr.* 2018;30(4):499-504.
9. Chinnadurai S, Sathe NA, Surawicz T. Laser treatment of infantile hemangioma: A systematic review. *Lasers Surg Med.* 2016;48(3):221-33.

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