Int J Acad Med Pharm, 2020; 2(3): 291-295
International Journal of Academic Medicine and Pharmacy
journal homepage: www.academicmed.org

Cutaneous Hemangioma: Our Experience Over 8 Years and Literature Review

Zeynep Bayramoglu[1*], Ayse Nur Ugur Kilinc [1], Yasar Unlu [1]

[1] Department of Pathology, Konya Training and Research Hospital, Konya, Turkey

Article info

Received: 14.05.2020
Received in revised form: 01.07.2020
Accepted: 04.08.2020
Available online: 05.09.2020

Keywords

Hemangioma
Benign vascular tumor
Angiosarcoma
Skin Tumor

Abstract

Hemangiomas are common benign vascular tumors. Clinical history, physical examination and imaging methods if necessary are generally used for the diagnosis. Excision and pathologic examination are performed if necessary. While most of the cases are treated without any problems it is important that some cases are malignant and that some cases are associated with syndromes. A total of 296 patients who were diagnosed with cutaneous hemangioma at our hospital between 2010 and 2018 were included in the study. Out of 296 patients in total, 152 were female and 144 were male. Of these patients, 128 were diagnosed with lobular capillary hemangioma, 41 with sinusoidal hemangioma, 3 with epithelioid hemangioma, 1 with microvenular hemangioma, 1 with hobnail hemangioma, 1 with angiokeratoma, 1 with spindle cell hemangioma, and 1 with tufted hemangioma. As 119 patients underwent only incisional biopsy no subtyping could be done. Hemangiomas are common benign vascular tumors with 14 different subtypes. We analysed our 296 patients with rare subtypes with literature review.

.

Research Article

INTRODUCTION

In the last edition of WHO classification of skin tumors (4th Edition, 2018), hemangiomas were divided into 14 subgroups as cherry hemangioma, sinusoidal hemangioma, microvenular hemangioma, hobnail hemangioma, glomeruloid hemangioma, spindle cell hemangioma, epithelioid hemangioma, tufted hemangioma, angiokeratoma, infantile hemangioma, congenital non-progressive hemangioma, rapidly involuting congenital hemangioma and non-involuting congenital hemangioma, lobular capillary hemangioma, verrucous venous malformation and arteriovenous malformation (Table-1). All of these hemangiomas are benign (ICD-O code is 0) in the International Classification of Diseases for Oncology (ICD-O)[1].

Table 1. Hemangioma subtypes in the last edition of WHO classification of skin tumors.

1)Cherry Haemangioma
2)Sinusoidal Haemangioma
3)Microvenular Haemangioma
4)Hobnail Haemangioma
5)Glomeruloid Haemangioma
6)Spindle Cell Haemangioma
7)Epithelioid Haemangioma
8)Tufted Haemangioma
9)Angiokeratoma
10)Infantile Haemangioma
11)Congenital Non-Progressive Haemangioma Rapidly Involuting Congenital Haemangioma and Non-Involuting Congenital Haemangioma
12)Lobular Capillary Haemangioma
13)Verrucous Venous Malformation
14)Arteriovenous Malformation

Cherry hemangioma is generally seen in adults at late middle ages and commonly located in the trunk and upper extremity as multiple, bright red, and asymptomatic papillary. In histopathological examination, well-demarcated, congested and dilated vessel proliferations that form groups in superficial dermis are seen[1,2]. Sinusoidal hemangioma is generally seen in the middle ages and more common in women. Although it is commonly located in the trunk it may also be seen in the breast and subcutaneous tissues of the extremities. In histopathological examination, it consists of lobulated, well-demarcated, irregular, dilated and congested thin-walled vessels. Microvenular hemangioma is a benign vascular lesion that is rarely seen in the young generally[1-3]. In histopathological examination, irregular capillaries and venules with small lumen are monitored in collagenized stroma. Atypia is not observed in endothelial cells.

Hobnail hemangioma is the wedge-shaped benign proliferation of intravascular papillary and hobnail endothelial cells in dermis and generally seen in adults and more commonly in the lower extremity in men. In typical macroscopic examination, a pale area is seen around the lesion. In immunohistochemical analysis, positivity with lesion D2-40 and CD31 was recorded. The lesion was also revealed to have lymphatic origin due to its D2-40 positivity. In

Corresponding author: Zeynep Bayramoğlu, E-Mail; drzeynepbayramoglu@hotmail.com , ORCID; 0000-0001-7075-8819
http://dx.doi.org/10.29228/jamp.45811

histopathological examination, a biphasic image occurs due to the different appearance of superficial and deep veins. Enlarged hobnail cells with prominent endothelial cells are monitored in superficial dermis. Vein structures with thin-narrow lumen are monitored in the deep areas of the lesion[4]. Glomeruloid hemangioma is a very rare lesion. It is associated with multicentric Castelman's disease and POEMS syndrome[5]. In histopathological examination, dilated vascular canals resembling glomerulus are seen in dermis. Although spindle cell hemangiomas are seen at any age they are seen in extremities especially in young adults. In histopathological examination, two components are noteworthy: well-demarcated, dermally-subcutaneously located, and large cavernous cavities and cellular spindle-epithelioid cells. The rates of these components may differ in the lesion. Epithelioid hemangioma is a benign vascular lesion generally smaller than 2 cm, most commonly seen in the 3rd and 5th decades and more common in women. In histopathological examination, well-developed vessel structures lined by epithelioid endothelial cells and stromal eosinophil leukocytes and lymphocytes are monitored. Endothelial cells are prominently monitored. Cytoplasmic vacuolization is sometimes observed[1-6]. Tufted hemangiomas are sporadic lesions that are very rare. They are common in children and young adults. In histopathological examination, capillary-type small vessels extending to the fat tissue in dermis form niches in lobular pattern[7]. Angiokeratoma generally appears on the dorsa. Angiokeratoma clinicopathologically has 4 types: Angiokeratoma Corporis Diffusum, Angiokeratoma of Fordyce, Angiokeratoma of Mibelli and Solitary Angiokeratoma. Infantile hemangioma is an infantile lesion mostly seen sporadically. In histopathological examination, it differs according to the growth phase of the lesion[1]. The tumor in proliferative phase consists of packed lobules and pericytes of capillaries lined by plump endothelial cells. With the maturation of the lesion, capillary dilatation, flattening of endothelial cells and dilatation of vessel lumens are seen. Congenital non-progressive hemangiomas are rare lesions and generally seen at birth and commonly appear in the head area.

Lobular capillary hemangiomas, also called pyogenic granulomas, are solitary, erythematous and bleeding papules or nodules that are rapidly growing, that show polypoid or peduncle development and that are more common in children and the young. They are generally smaller than 2 cm. In histopathological examination, lobulated development of dermally located capillaries is monitored[8]. Verrucous venous malformation is a rare congenital lesion. These lesions generally appear on the lower extremity. In histopathological examination, acanthosis, papillomatosis, hyperkeratosis, and parakeratosis are monitored in epidermis and crusting overlies an expanded papillary dermis with numerous dilated and congested capillaries and venules[9]. Arteriovenous hemangiomas are solitary papillary lesions that are commonly located in the head and neck area and their size are mean 0.5-1 cm. In histopathological examination, they are characterized by irregular proliferation of arterial, venous or capillary vessels lined by well-demarcated, thick and thin walled in fibrous or fibromyxoid area, and single-row endothelial cells in superficial and mid dermis[1]. In the basis of the lesion, there is an artery feeding the lesion. In addition, arteriovenous shunts may be monitored.

MATERIAL and METHOD

Ethic approval

This study was approved by the Ethical Committee in Konya Training and Research Hospital, Turkey (02 July 2020, 48929119/774)

Patients and study design

A total of 296 patients who were diagnosed with cutaneous hemangioma in their incisional and excisional biopsy at our hospital between 2010 and 2018 were included in the study. The patients were scanned in the information system of the hospital and their demographic and clinical data were evaluated. Preparations with hematoxylin-eosin staining were obtained from our archive and re-evaluated. This study was approved by the Ethical Committee in Konya Training and Research Hospital, Turkey.

RESULTS

A total of 296 patients were included in the study; 152 were female and 144 were male. Mean age of the patients was 39.2. Thirty-one patients were at the ages between 0-18. Out of 296 patients, 128 were diagnosed with lobular capillary hemangioma, 41 with sinusoidal hemangioma, 3 with epithelioid hemangioma (Figure-1), 1 with microvenular hemangioma(Figure-2), 1 with hobnail hemangioma (Figure-3), 1 with angiokeratoma, 1 with spindle cell hemangioma(Figure-4), and 1 with tufted

hemangioma (Figure-5). As 119 patients underwent only incisional biopsy no subtyping could be done. When the locations of the lesions were evaluated it was observed that they were located in the head and neck area of 144 patients, in the upper extremity of 86 patients, in the trunk of 34 patients, in the lower extremity of 24 patients and in genital area of 12 patients. When the patients who underwent excisional biopsy were evaluated the largest hemangioma diameter was recorded as 2.2 cm and the smallest was 0.1 cm. Mean hemangioma diameter was 0.84 cm.

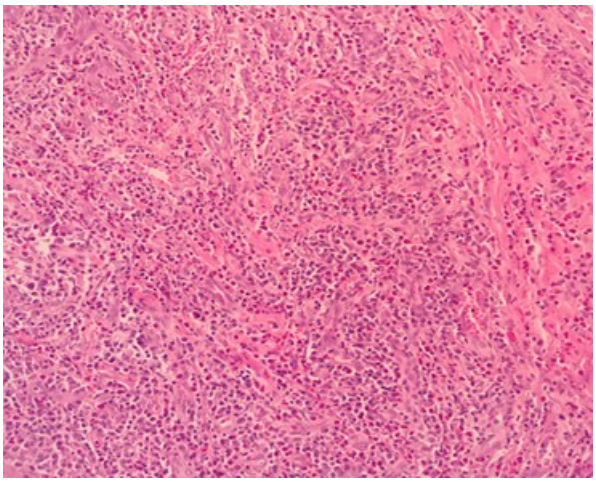

Figure 1. Epithelioid Haemangioma (H&E 200x)

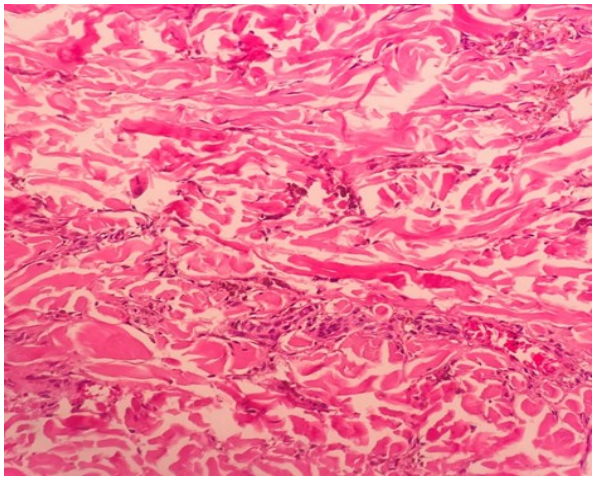

Figure 2. Microvenular Haemangioma (H&E 400x)

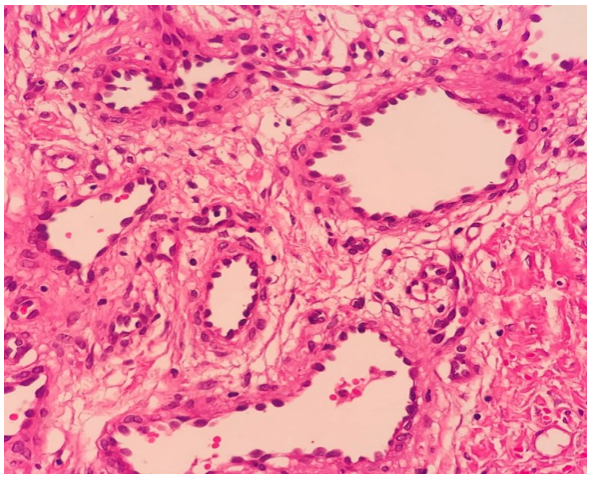

Figure 3. Hobnail Haemangioma (H&E 400x)

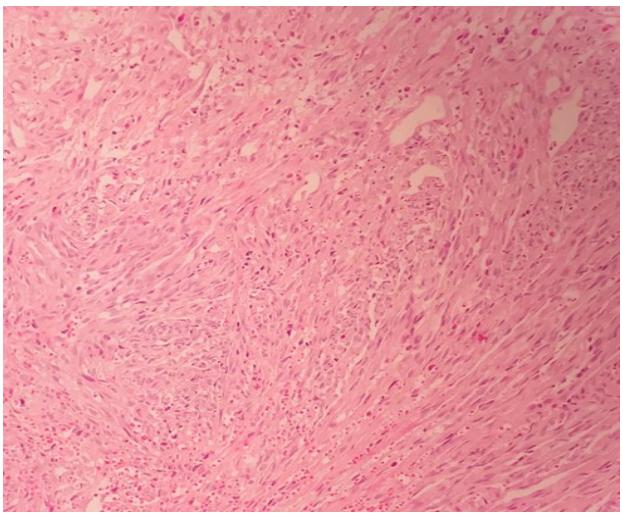

Figure 4. Spindle Cell Haemangioma (H&E 100x)

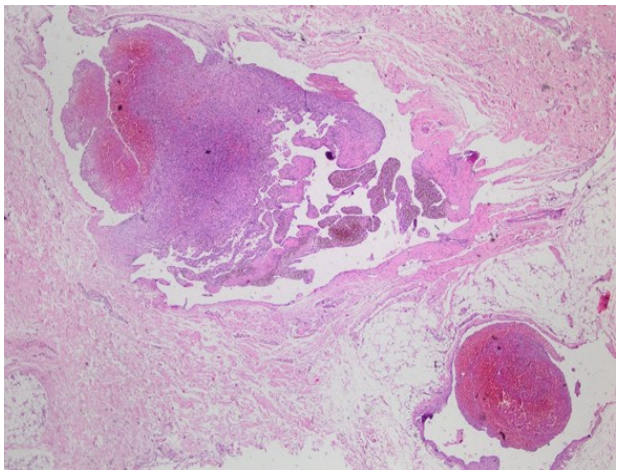

Figure 5. Tufted Haemangioma (H&E 20x)

In patients who were diagnosed with lobular capillary hemangioma, the lesion was located in the upper extremity of 68 patients, in the lower extremity of 32 patients, in the head and neck area of 26 patients and in genital area of 2 patients. In patients who were diagnosed with sinusoidal hemangioma, the lesion was located in the trunk of 37 patients, in the upper extremity of 2 patients and in the lower extremity of 2 patients. In 3 patients who were diagnosed with epithelioid hemangioma, the lesion was located in the lower extremity of 1 patient, in the head and neck area of 1 patient and in the genital area of 1 patient. The lesion was located in the upper extremity of 1 patient who was diagnosed with microvenular hemangioma, in the head and neck area of the patient who was diagnosed with hobnail hemangioma, in the trunk of the patient who was diagnosed with intramuscular hemangioma, in the lower extremity of the patient who was diagnosed with spindle cell hemangioma and in the trunk of the patient who was diagnosed with tufted hemangioma.

DISCUSSION

Hemangiomas are the lesions we commonly encounter in our daily life, but they have a broad differential diagnosis. As its differential diagnosis list involves many entities from malignancies to reactive processes it is a group that we need to be very careful about. In the last edition of WHO classification of skin tumors (4th edition, 2018), hemangiomas were re-classified and the reclassification was mentioned in the introduction part[1].

While evaluating hemangiomas growth pattern, low mitotic activity, arrangement of endothelium in a single layer and maturation in the lesion generally suggest a benign diagnosis. However, none of the histopathological features are sufficient alone. Immunohistochemistry may be utilized in patients when we are irresolute for differential diagnosis of hemangiomas. In immunohistochemical examination, CD34, CD31, Factor 7 and ERG can be used as endothelial markers. Smooth muscle actin can be used for pericyte cells. Angiosarcoma should definitely be considered in differential diagnosis while evaluating hemangiomas.[10] Angiosarcomas are generally seen among collagen bundles in reticular dermis and among irregular, demarcated and infiltrative vessel cavities that reveal dissecting growth pattern in the subcutaneous fat tissue. Endothelial cells have large and hyperchromatic nuclei in angiosarcomas. In addition, stratification and cytoplasmic vacuolization in endothelial cells are important findings for angiosarcoma[11].

When we reviewed literature lobular capillary hemangioma was the most common among our patients, which was in compliance with literature. Although lobular capillary hemangioma is commonly seen in the finger and head and neck area it has been reported to also appear on periocular area, on lip, inside the bone and on intraabdominal area[8,12-14]. In addition, lobular capillary hemangiomas are rarely congenital according to the records of literature. Only 2 of our patients had congenital lobular capillary hemangioma and 2 had lobular capillary hemangioma in the head and neck area.

Hemangiomas that are rarely seen were given in Table 2. Only 3 of our patients had epithelioid hemangioma and they were all female. In the evaluation of the locations, they were generally in the head and neck area. It was in the head and neck area in 1, in the trunk in 1 and in the lower extremity in 1 of our patients. It has been reported that they are also seen in the heart and tongue.[15,16] One of our male patients was diagnosed with microvenular hemangioma and the lesion was located in his upper extremity. A patient diagnosed with microvenular hemangioma was examined in terms of PR immunoreactivity due to the history of hormone therapy and it was positive.[17] Therefore, it was stated that progesterone may be associated with microvenular hemangioma. Only 1 female patient had hobnail hemangioma in the head and neck area. Hobnail hemangioma is very rare and the largest series in literature consist of 6 patients. Three of these 6 patients had congenital hobnail hemangioma[4]. One male patient among our patients had angiokeratoma in his lower extremity. One male patient among our patients had spindle cell hemangioma and the lesion was located in his lower extremity. Although spindle cell hemangiomas are commonly located in the lower extremity there are also patients in whom the lesions are located in the spleen, bone, and upper lip or have intramedullary location[18-21]. Only 1 of our female patients had tufted hemangioma in her trunk. Tufted hemangioma is a very rare lesion. According to a recent study including 54 patients, tufted hemangioma was most commonly seen in the lower extremity, there was no difference between the genders and 45 patients had solitary while 9 had multiple hemangioma. In addition, immunochemical analysis with CD31, WT-1, D2-40, Prox and GLUT-1 was performed in that study in order to show the lymphatic differentiation and a diffuse positive reaction with CD31 and WT-1, focal positive reaction with D2-40 and Prox and negative reaction with GLUT-1 were recorded in proliferous endothelial cells. In conclusion, they stated in their study that there was lymphatic differentiation in tufted hemangiomas[7]. There was focal positive reaction with D2-40 in our patient, which confirms lymphatic differentiation.

Table 2. Summary of our rare hemangiomas.

	SUBTYPE	AGE	GENDER	LESİON DİAMETER	LOCALİZATİON
1	Epithelioid Haemangioma	25	Female	0,8CM	Lower Extremity
2	Epithelioid Haemangioma	42	Female	1,6CM	Head and Neck Area
3	Epithelioid Haemangioma	25	Female	0,8CM	Genital Area
4	Microvenular Haemangioma	40	Male	1,5CM	Upper Extremity
5	Hobnail Haemangioma	26	Female	0,5 CM	Head and Neck Area
6	Angiokeratoma	15	Male	2,5CM	Trunk
7	Spindle Cell Haemangioma	17	Male	0,6CM	Lower Extremity
8	Tufted Haemangioma	18	Female	1CM	Trunk

CONCLUSSION

In conclusion, hemangiomas are the lesions that we commonly encounter in our daily routine and their differential diagnosis may sometimes be hard. Its association with syndromes is also clinically very important. In this study, we evaluated our experience on hemangioma with its rarely seen subtypes with literature review and its new classification.

Conflict of interest

No conflict of interest was declared by the authors.

REFERENCES

1. Elder DE, Massi D, Scolyer RA, Willemze R. WHO classification of Skin tumors: an update based on the 2018 (4th) edition; 2018:346-55 p.
2. Destek S, Gul VO. Clinical and Pathological Evaluation of Benign Skin Lesions. *Sains Malaysiana.* 2019;48(12):2693-9.
3. Díaz-Flores L, Gutiérrez R, González-Gómez M, Sáez FJ, Díaz-Flores Jr L, Carrasco JL, et al. Sinusoidal hemangioma and intravascular papillary endothelial hyperplasia: interrelated processes that share a histogenetic piecemeal angiogenic mechanism. *Acta Histochemica*. 2018;120(3):255-62.
4. AbuHilal MD, Breslavet M, Ho N, Taylor G, Pope E. Hobnail hemangioma (superficial hemosiderotic lymphovascular malformation) in children: a series of 6 pediatric cases and review of the literature. *Journal of Cutaneous Medicine and Surgery.* 2016;20(3):216-20.
5. Aragüés IH, Pérez AP, Blanco CC, Blanco VP, Fernández RS. Glomeruloid Hemangioma and POEMS Syndrome. Actas Dermo-Sifiliográficas (English Edition). 2017;108(2):e15-e9.
6. Sedran L, Bonaso M, Mettus A, Roccia F. Epithelioid Hemangioma of the Face. *Journal of Craniofacial Surgery*. 2018;29(8):e736-e9.
7. Su X, Liu Y, Liu Y, Ma C. A retrospective study: Clinicopathological and immunohistochemical analysis of 54 cases of tufted angioma. *Indian Journal of Dermatology, Venereology, and Leprology.* 2020;86(1):24.
8. Chan C, Iv M, Fischbein N, Dahmoush H. Lobular capillary hemangioma of the mandible: A case report. *Clinical Imaging*. 2018;50:246-9.
9. Beijnen UE, Saldanha F, Ganske I, Upton J, Taghinia AH. Verrucous venous malformations of the hand. *Journal of Hand Surgery* (European Volume). 2019;44(8):850-5.
10. Johnson EF, Davis DM, Tollefson MM, Fritchie K, Gibson LE. Vascular tumors in infants: case report and review of clinical, histopathologic, and immunohistochemical characteristics of infantile hemangioma, pyogenic granuloma, noninvoluting congenital hemangioma, tufted angioma, and kaposiform hemangioendothelioma. *The American Journal of Dermatopathology*. 2018;40(4):231-9.
11. Moon SY, Eun DH, Jang YH, Lee S-J, Lee WJ. Primary angiosarcoma of the skin presenting as mild erythema. *Annals of Dermatology*. 2017;29(6):809-10.
12. Heinz P, Hoffmann M, Hosie S. Bleeding to the point of anemia in lobular capillary hemangioma of the upper lip. *Deutsches Ärzteblatt International*. 2019;116(8):134.
13. Stagner AM, Jakobiec FA. A critical analysis of eleven periocular lobular capillary hemangiomas in adults. *American Journal of Ophthalmology*. 2016;165:164-73.
14. Hashimoto S, Obinata D, Yamaguchi K, Sakurai F, Yoshida T, Yoshizawa T, et al. Case of caval lobular capillary hemangioma mimicking tumor thrombus. IJU Case Reports. 2019;2(2):80-2.
15. Motwani M, Karemore T, Sarate KO. Epithelioid hemangioma: A rare entity to report. *Journal of Indian Academy of Oral Medicine and Radiology*. 2018;30(3):321.
16. Machado I, Chong A, Serrano A, Naranjo Ugalde AM, Pineda D, Savón L, et al. Epithelioid hemangioma (angiolymphoid hyperplasia with eosinophilia) of the heart with peripheral eosinophilia and nephrotic syndrome. *International Journal of Surgical Pathology*. 2016;24(1):59-65.
17. Juan YC, Chen CJ, Hsiao CH, Chiu TM. A microvenular hemangioma with a rare expression of progesterone receptor immunocreativity and a review of the literature. *Journal of Cutaneous Pathology*. 2018;45(11):847-50.
18. Nasser R, Ashayeri K, Legatt AD, Houten JK. Intramedullary spindle cell hemangioma: case report. *Journal of Neurosurgery: Spine*. 2016;25(3):379-82.
19. Keil F, Dietmaier W, Hoffstetter P, Hillmann A, Evert M. ZFP36-FOSB Fusion in a Hemorrhagic Epithelioid and Spindle Cell Hemangioma of Bone: Is there a family of FOSB-rearranged vascular neoplasms of the bone? *Histopathology*. 2019.
20. Murakami K, Yamamoto K, Sugiura T, Kirita T. Spindle Cell Hemangioma in the Mucosa of the Upper Lip: A Case Report and Review of the Literature. Case reports in dentistry. 2018;2018.
21. Gao B-Q, Zhou D-K, Qian X-H, Zhang W, Ying L-X, Wang W-L. Spindle cell hemangioma of the spleen: A case report. *Medicine*. 2019;98(9).