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Arterio-Venous Hemangioma of Left Thigh

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ABSTRACT

Arteriovenous hemangioma is a distinct benign, acquired vascular tumor. The recognition of this entity on various diagnostic modality and treatment can be challenging. A 14 year male patient presented with complaints of gradually increasing swelling of left thigh since 6 months. Clinically it was suspected of soft tissue tumor. On radioimaging MRI left thigh and contrast shows 5.9 x 2.1x2.0 cm size, well-defined, multi loculated, cystic lesion with septae. On histopathology reported as arteriovenous hemangioma of left thigh. The skeletal muscle hemangiomas are uncommon soft tissue tumors. We are presenting this case for its rarity clinical, radiological and histopathological findings.

Keywords: Vascular tumor, Hemangioma, Arteriovenous malformation.

INTRODUCTION

Arteriovenous hemangioma (AVH) is a distinct benign, acquired vascular tumor. On histopathology AVH shows thick and thin-walled sized arteries and veins in close association with one another. [1,2] The incidence of AVH is noted infrequently in the literature. They have unpredictable behavior and a high recurrence rate. Appropriate diagnosis and adequate line of treatment will be required to manage these cases. In our case it was presented as deep soft tissue mass lesion. On clinical, radiological and histopathological findings case was evaluated.

CASE REPORT

A 14 year old male patient presented with complaints of gradually increasing swelling of left thigh since 6 months. There was history of pain off and on which increased in last decades. On clinical examination, showed a single, large soft tissue swelling on left thigh measuring 6 x 3 x 2 cm. The margins were regular. It was soft to firm in consistency, pulsetile and movable. There was no any regnional lymphadenopathy. There was no any contributory past/family history. The USG of left thigh swelling showed 6.2 x 5.2 x 1.7 size, a well defined hyper-echoic lesion. The lesion shows multiple torturous, anechoic vascular channels. The lesion was intramuscular and within lesion shows small thrombotic areas. On USG reported as suggestive of AV malformation.

On MRI left thigh and contrast showed 5.9 x 2.0 cm size, well-defined, multi loculated, cystic lesion with septae seen in the anterio lateral aspect of left distal

femur extending in the vastus intermedius muscle. The lesion appeared predominantly T2 hyper intense and iso intense on T1 Wt images. There were hypo-intensities within the lesion. The lesion revealed heterogenous enhancements on contrast studies. The lesion was in close contact with cortex of distal femur. There was no bony destruction. On MRI reported as? Hemangioma,? lymphatic malformation? Nerve sheath tumor. The wide surgical excision was done. The excised specimen was sent for histopathological evaluation.

Figure- 1. Specimen of excised left thigh swelling dark brown, solid, cystic areas filled with blood.

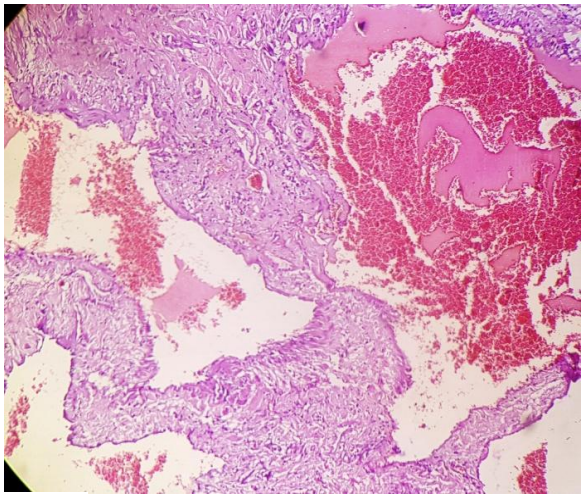

Figure-2. Photomicrograph showing numerous small and large size thick- and thin-walled dilated interconnecting vascular spaces filled with blood.(H & E stain,100 x)

We received a specimen of excised left thigh swelling, on gross morphological examination; it was totally measuring 5.7 x 3.6 x 2.2 cm. The lesion was soft to firm. On cut section showed dark grey brown solid areas with multiple variable sized cystic areas filled with blood (Figure 1). On histopathology, serial sections showed numerous small and large size thick- and thin-walled dilated interconnecting vascular spaces of arteries and veins lined by endothelium and lumen is filled with blood (Figure 2, 3). The vessels are in close association with one another. The features of recanalization was noted. The intra-lesional nerves were absent. In our study, we performed histochemical stains Masson's trachoma, and Verhoeff-van Gieson's for elastic tissue which showed weak staining. The areas of hemorrhage, thrombotic vessels and myxoid change are noted. The surrounding areas showed fibromuscular tissue with congestion. On histopathology reported as arterio-venous hemangioma along with areas of hemorrhage inflammation, fibrosis, myxoid change and few thrombotic vessels. The wide surgical excision was done. The patient had an uneventful post-operative period and advised regular follow up.

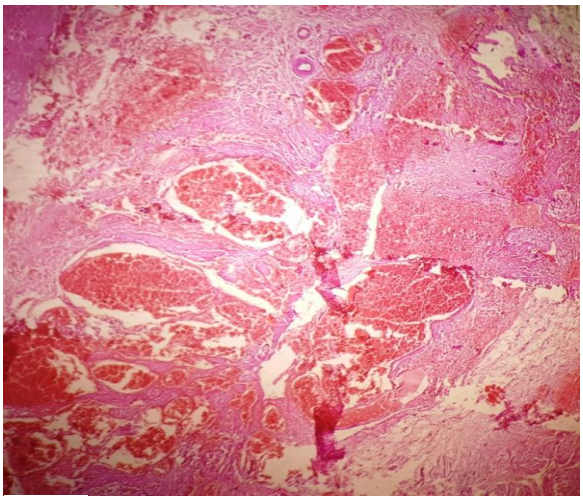

Figure-3. Photomicrograph showing numerous small and large size thick- and thin-walled dilated vascular spaces filled with blood.(H & E stain,40x)

DISCUSSION

We have a case of deep situated intramuscular hemangioma. Intramuscular hemangiomas are rare benign tumors, making up 0.8% of all hemangiomas [3] Arteriovenous hemangioma (AVH) is a distinct benign, acquired vascular tumor. Its main histological feature is multiple thick-

and thin-walled vascular spaces resembling arteries and veins, respectively. Absence of internal elastic lamina in thick-walled blood vessels suggests ectasic veins. The incidence of AVH is noted infrequently in the literature. The AVH represent a histologic variant of hemangiomas and it is regarded by some as an Arteriovenous Malformation (AVM) causing varying degrees of shunting.[4] Hemangiomas are vascular tumors that are rarely apparent at birth, grow rapidly during the first 6 months of life, involute with time and do not necessarily infiltrate but can sometimes be destructive. Vascular malformations are irregular vascular networks defined by their particular blood vessel type, present at birth, slow growing, early small, later expansion, infiltrative, and destructive.

The etiology of AVH is uncertain. The various causes are developmental, endocrine, reactive and inflammatory. [5] In our case there was no any contributory findings were noted. The hemangiomas are divided into various types - cavernous, capillary, venous, arteriovenous, epitheloid, spindle cell, lobular capillary etc. [6]

In our case location was deep in the thigh muscle.AVH are benign vascular tumor divided into types according to the depth of involvement: a) Deep form b) Superficial form. The deeply situated types of AVH are mostly noted at the head, neck and lower extremity in young adults. These are associated with arteriovenous shunting and soft tissue hypertrophy. The malignant vascular tumor angiosarcoma of deep soft tissue is a rare. [7] While superficial AVH also named as cirsoid aneurysm or acral arteriovenous tumor are commonly noted in elderly adults and mostly at the head and neck region.

On histopathology AVH shows thick and thin-walled sized arteries and veins in close association with one another. Focally, some tumors resemble capillary and cavernous hemangioma. There may be focal thrombosis, secondary dystrophic calcification and mild inflammation. During grossing of these lesions the serial sections are helpful in demonstrating continuities or shunts between arteries and veins.

In vascular malformations shows dysplastic vessels with characteristic features such as leaky vessels, eccentric intimal fibrosis of artery, arterializations of vein, and disrupted internal elastic lamina. Use of elastic tissue stains are useful ancillary tools to distinguish between AVM and hemangiomas. The histochemical stains such as Verhoeff-van Gieson (VVG), Masson's trichrome (MT), and Toluidine blue can be used. [8] The arteries and arterioles with elastic lamina in their walls are an integral part of an AVM. The triggering events like sepsis, trauma, puberty, and pregnancy may increase the lesion size. In situation where the diagnosis is not appropriate, an AVM may result in ulceration, hemorrhage, and wide surgical resection.

On Haematoxyline and Eosin stained sections the presence of the intra-lesional nerves is useful to distinguish between AVM and hemangiomas .In the study by Pawane P et al showed that the presence of intra-lesional nerves in 81.8% cases of AVM while they were seen in only 6% in hemangiomas. [8,,9] In our case nerves were absent.

Hemangioma and vascular malformation, especially AVM, should be clearly differentiated to reduce the risk of treatment failure and recurrence. [4] They have unpredictable behavior and a high recurrence rate. To avoid misdiagnosis must perform a complete vascular update study on recent developments in the diagnosis, management, and pathogenesis of vascular anomalies. So it will be helpful to determine the magnitude, flow, and extent of the lesion.

This case was treated with wide excision. It is important to do vascular work up for the conservative and surgical treatment options. Corticosteroids, interferon, and vincristine have been successfully used. The surgical management involves excision, laser treatment or both.

The multidisciplinary approach is frequently necessary in managing these lesions. Almost all vascular malformations and nearly 40% of hemangiomas eventually require intervention. [10]

CONCLUSION

Arteriovenous hemangioma is a distinct benign, acquired vascular tumor. The recognition of this entity on various diagnostic modality and treatment play important role. We are presenting this case for its rarity clinical, radiological and histopathological findings.

Conflict of interest - no
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REFERENCES

1. Garzon MC, Enjolras O, Frieden IJ. Vascular tumors and vascular malformations: evidence for an association. J Am Acad Dermatol. 2000 ;42(2 Pt 1):275-9.
2. Chang MW. Updated classification of hemangiomas and other vascular anomalies. Lymphat Res Biol. 2003;1(4):259-65.
3. Wild AT, Raab P, Krauspe R. Hemangioma of skeletal muscle. Arch Orthop Trauma Surg. 2000;120(3-4):139-43
4. EnzingerF.M., Weiss.W. Benign tumors and tumorlike lesions of blood vessels. In: Soft tissue tumors, 1995, 3rd edition, pg. 588-593.
5. Koutlas IG, Jessurun J. Arteriovenous hemangioma : a clinicopathological and immunohistochemical study. J Cutan Pathol. 1994 ;21(4):343-9.
6. Bruder E, Perez-Atayde AR, Jundt G, Alomari AI, Rischewski J, Fishman SJ et al. Vascular lesions of bone in children, adolescents, and young adults. A clinicopathologic reappraisal and application of the ISSVA classification. Virchows Arch. 2009 ;454(2):161-79.
7. Jagtap SV, Kumar S, Kulkarni SR et. al. Angiosarcoma of deep soft tissue presented as mass lower leg. Int J Health Sci Res. 2014;4(11):274-277.
8. Pawane P, Anshu, Gangane N. Hemangiomas versus arterio-venous malformations: Role of elastic stains and mast cell density. Indian J Pathol Microbiol 2014;57:191-5
9. Adegboyega PA, Qiu S. Hemangioma versus vascular malformation: presence of nerve bundle is a diagnostic clue for vascular malformation. Arch Pathol Lab Med. 2005 ;129(6):772-5.
10. Richter GT, Friedman AB. Hemangiomas and vascular malformations: Current theory and management. Int J Pediatr .2012; 2012:645-678.

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