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BROAD LIGAMENT ANGIOMYOMAS: RARE SERIES OF TWO CASES AND A MINI REVIEW

Histopathology

Dr. Bhavna Sharma* MD, DNB Pathology, Assistant Professor, Department Of Pathology, Dr. Baba Saheb Ambedkar Hospital , New Delhi *Corresponding Author

Dr. Nidhi Mahajan MD, DNB Pathology, Assistant Professor, Department Of Pathology, Dr. Baba Saheb Ambedkar Hospital , New Delhi

Dr.Karsing Patiri MD Pathology, Associate Professor And Head Of Unit, Department Of Pathology, Dr. Baba Saheb Ambedkar Hospital , New Delhi

Dr. Khushboo Dewan MD Pathology, Assistant Professor, Department Of Pathology, Dr. Baba Saheb Ambedkar Hospital, New Delhi

Dr.Shyam Lata Jain MD Pathology, Professor, Department Of Pathology, Dr. Baba Saheb Ambedkar Hospital, New Delhi

ABSTRACT

Angiomyomas are benign, small sized smooth muscle tumors which arise in the subcutis of extremities. Broad ligament being the primary site of these tumors along with large size, both are extremely rare. We hereby report not one, but two such rare cases alongwith a mini review of the same. This will add to the existing knowledge of this tumor.

KEYWORDS

Smooth muscle, angiomyoma, broad ligament

INTRODUCTION

Angiomyomas are benign, small sized smooth muscle tumors which arise in the subcutis of extremities. Broad ligament being the primary site of these tumors along with large size, both are extremely rare. We hereby report not one, but two cases alongwith a mini review of the same.

Case Report

Case 1: A 46 year female presented to surgery OPD with a right inguinal swelling and pain since 1 year. Hernia was suspected and ultrasonography confirmed the same. However, intraoperatively an encapsulated circumscribed mass was identified arising from the round ligament. Excision was done and tissue was sent for histopathology with a clinical diagnosis of lipoma. Grossly, mass was encapsulated measuring 10X6X4 cm. On serial slicing it was homogenous white with areas of myxoid change.[Fig 1A, 1B] Microscopy revealed bundles of smooth muscle cells punctuated by thick walled vessels. The inner layer of the smooth muscle of the vessel were arranged in an orderly cicumferential fashion and the outer layer was found to spin away from the vessel wall merging with the less ordered peripheral muscle fibres.[Fig 2A] Large areas of myxoid change and hyalinisation were also noted.[Fig 2B] These were found to be diffusely immunopositive for SMA [Fig 2C] and Demin and negative for HMB-45.[Fig 2D] Ki-67 was low.

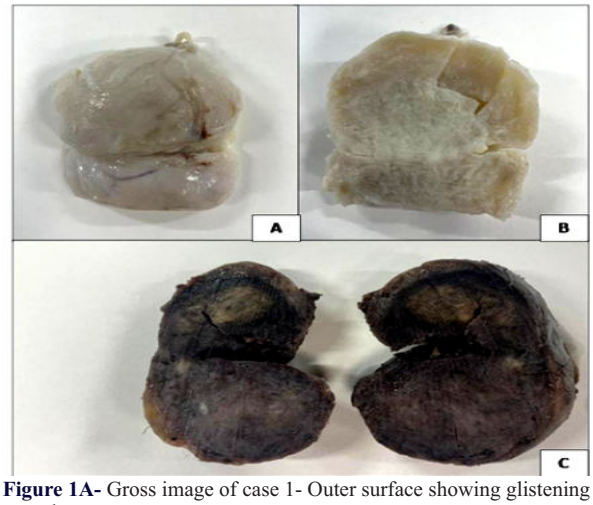

Figure 1A- Gross image of case 1- Outer surface showing glistening capsule

Figure 1B- Cut surface of case 1- showing whorling along with myxoid change

Figure 1C- Gross image of case 2 showing encapsulated tumor with markedly congested surface

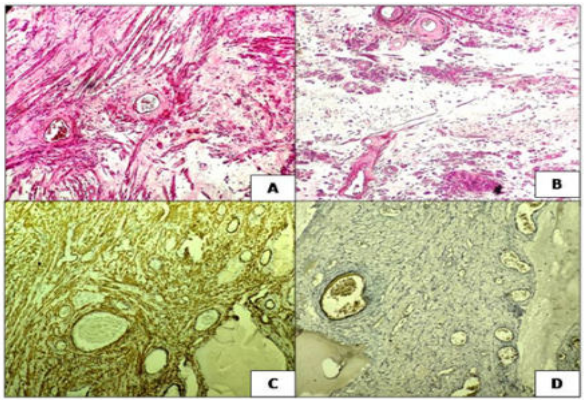

Figure 2A- Smooth muscle punctuated by thick walled vessels with outer layer radiating away from the vessel wall

2B- Areas of myxoid change

2C- Diffuse Smooth Muscle Actin positivity

2D- Negativity for HMB-45

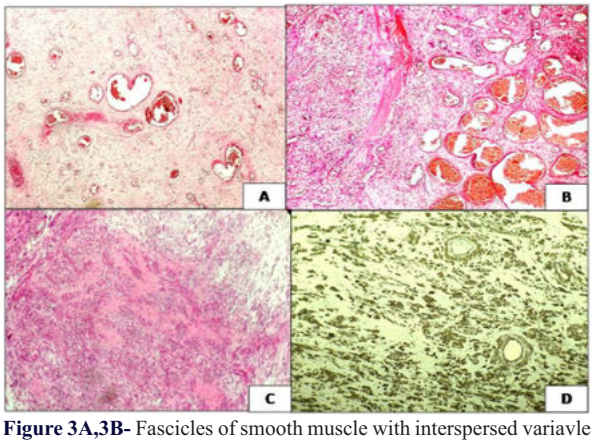

Figure 3A,3B- Fascicles of smooth muscle with interspersed variavle sized vascular channels

Figure 3C- Areas of hyalinisation

Figure 3D- Diffuse Smooth Muscle Actin positivity

Case 2: A 39 year old pregnant female, 32 weeks POG came to Gynae OPD with complaints of decreased fetal movements. Ultrasonography revealed oligohydramnios along with growth restriction. In addition, left sided broad ligament mass was identified with a differential of a cyst or degenerated fibroid. Elective caesarean section was done and the broad ligament mass was also excised and sent for histopathology. Grossly the mass was encapsulated, globular, measuring 9X6X4 cm. Serial slicing revealed areas of congestion.[Fig 1C] On microscopy, sections examined showed a well encapsulated moderately cellular lesion comprising of variably sized vascular channels interspersed between sweeping fascicles of spindle cells and areas of hyalinisation.[Fig 3A, 3B, 3C] No nuclear pleomorphism/ mitoses/necrosis were seen. The tumor cells were diffusely immunopositive for SMA.[Fig 3D]

DISCUSSION

Cutaneous or superficial leiomyomas are of two types: pilar and genital. Angiomyomas alongwith cutaneous leiomyomas were previously called Tuberculum dolorosum due to its pain producing characteristics.[1]But later, these were differentiated from cutaneous leiomyomas as origin from vascular smooth muscle was suggested and hence the name Vascular Leiomyoma.[1]Their rarity is exemplified by the fact that these comprise only about 5% of all benign soft tissue tumors. [2]In the series by Hachisuga et al, majority of these arose from lower extremities followed by upper extremities and very few on head and trunk.[2]Uterine angiomyomas are also encountered but are rare.[3]In the current case the site of origin was broad ligament which is even rarer than uterine origin .[4] Very few case reports of broad ligament angiomyomas exist in literature.[5-10] Not only the location, the size was also huge, as compared to the <2 cm under usual circumstances.[2]We report these cases due to rarity of the site as well as size.

Etiologic factors include trauma, effect of hormones and venous stasis. Some postulate that these are actually arteriovenous malformations. But this is still a matter of debate.[11]

Pain is the characteristic presentation. it is postulated that contraction of smooth muscle leads to transient ischemia and hence pain.[12] Another hypothesis attributes pain to presence of intratumoral nerve fibres.[13]Most of the previously described cases and our cases too presented with pain.[Table 1]

Table 1- Review Of Broad Ligament Angiomyomas

Author	Presenting Complaint	Age	Size	Histologic type
Hemalatha et al	Mass per vaginum since 6 months	40 years	10x6x5cm	Capillary
S Agarwal et al	Abdominal lump and pain since 2 months	45 years	25x10x10cm	Venous
Guven et al	Abdominal Pain since 2 years	43 years	12x10 cm	Capillary
Mane et al	Abdominal pain and mass	42 years	Multiple, size -NA	Capillary
Maheshwari et al	Pain and abdominal mass	45 years	5.5x4x3.5cm	Venous
Cobellis et al	Pain since 1 year	52 years	15x15 cm	Capillary

Histologically, angiomyomas are of three types: capillary, venous and cavernous.[12]Both our cases were of the venous types. In the previously reported broad ligament angiomyomas, majority are of capillary type, very few of venous type have been reported. {Table 1} Angiomyomas can show areas of hyalinisation and myxoid changes, as were evident in our case too.[12] Even areas of calcification and fat may be seen. Differentialdiagnoses broadly includes leiomyoma, vascular tumors, angiomyolipoma , myopericytoma , angiofibroma , angiomyofibroblastoma and sometimes schwannoma.Approach to differentiate amongst these is outlined briefly in Table 2. Both the cases were immunopositive for smooth muscle actin and thusthe diagnosis was confirmed .

As described above, mostly Angiomyomas are less than 2 cm. But as can be seen in Table 1, most of the broad ligament angiomyomas described are >2 cm. The inaccessible location may lead to delay in presentation and diagnosis and hence can be put forward as a possible explanation to larger sizes of broad ligament Angiomyomas.

Differential Diagnosis	Histology	Immunohistochemistry
Leiomyoma	Bundles of smooth muscle, no intervening vessels	SMA+, CD34 -
Vascular tumors	Only vessels, no smooth muscle	CD34+, SMA-
Angiomyolipoma	Smooth muscle, vessels and fat	HMB-45+, SMA+, CD34+
Angiofibroma	Genital location, a cellular spindle cell component arranged randomly or in short fascicles, vague palisades, or swirls around a prominent vasculature, +/- mature fat	CD34+ strong and diffuse,ER+, PR+
Angiomyofibroblastoma	Genital location, Ectatic vessels surrounded by clusters of epithelioid or spindle cells, some of which blend or fan out from the muscular wall of vessels	Vimentin+, Desmin+, SMA-, CD34-
Myopericytoma	Oval to short fusiform cells that demonstrate a striking multilayered concentric growth around the vessels, which may appear small and rounded or elongated and ectatic	SMA +, Most difficult to differentiate from Angiomyoma
Schwannoma	No muscle, large hyalinised vessels, areas of myxoid change	S-100+, SMA-, CD34-

Treatment is complete surgical excision. Recurrences are extremely rare.

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