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Angiomyolipoma of Cervix: Case report of a rare entity

[1]Dr. Shrushti Mishra, [2]Dr. Nikita R. Vijay, [3]Dr. Sulbha Joshi

[1-3]Department of Obstetrics and Gynecology, NKP Salve Institute of Medical Science, Nagpur, Maharashtra, India

Corresponding Author: Dr. Nikita R. Vijay Department of Obstetrics and Gynecology, NKP Salve Institute of Medical Sciences, Nagpur, Maharashtra, India

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Abstract

Angiomyolipoma (AML) is a benign Mesenchymal tumor which occurs most frequently in Kidney and is commonly associated with Tuberous Sclerosis. Histological features are characterized by admixture of Blood vessel, smooth muscle cells and adipose tissue. Cervical angiomyolipomas are rare and a very few cases have been reported in literature. We report a case of Cervical angiomyolipoma without any features of Tuberous sclerosis. Final diagnosis of Angiomyolipoma was made Postoperatively after Histopathology examination. Cervical angiomyolipoma though rare tumor but should be considered as a differential diagnosis for lower abdominal mass and Abnormal uterine bleeding.

Keywords: Angiomyolipoma, PEComa, Cervical Fibroid, Abnormal uterine bleeding.

Introduction

Angiomyolipoma (AML) is a Benign Mesenchymal tumor composed of vascular cells, smooth muscle cells and adipose tissue, belonging to family of Perivascular epithelioid cells tumors (PEComa).1 PEComa is designated as "mesenchymal tumor composed of histologically and immunohistochemically distinctive perivascular epitheloid cells". PEComa is generally acknowledged as a family of tumor which include Renal or extrarenal angiomyolipoma(AML), clear cell "sugar" tumor of lungs(CCST), lymphangioleiomyomatosis (LAM), clear-cell myomelanocytic tumor of falciform ligament, and group of rare and clear cell tumor of different locations.[2,3] Angiomylipoma is usually associated with Tuberous Sclerosis. Most common site of it being kidney and other sites include Liver, Retroperitoneum, Large Bowel, Nasal cavity and Pelvic region. Angiomylipoma of Cervix without any association with Kidney is a rare entity and very few cases have been reported in literature.[4,5]

Case Report

A 48years old Female Para3,Living3, last child birth 22 years back with previous cycles being regular with average flow, came with complaint of mass coming out of vagina since 1year. On examination her vitals were stable. On Per-abdomen examination abdomen was soft, non tender, no mass felt. Per-speculum examination showed mass of size 10 x 7cm, pink, globular, firm with ulcerative lakes and eroded surface arising from anterior lip of cervix, hanging outside

vagina. No Cystocele, No Rectocele. On Per-vaginal examination Uterus was bulky, bilateral fornices free. On Per-rectal examination Cervix felt anteriorly, Rectal mucosa free. Fig 1

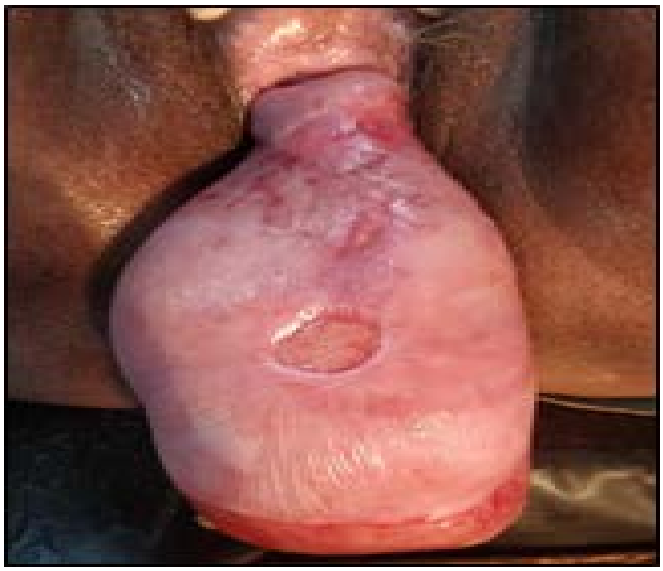

Figure 1: Clinical picture of mass descending per vagina.

Patient underwent Mass excision, Intra operative finding were mass of 10 x 7cm globular arising from the anterior lip of cervix. On gross examination single, globular greyish white tissue tumor mass of size 10.2 x 7 x 5cm (Figure 2A). Cut surface is homogenous white with few haemorrhagic areas. (Figure 2B).

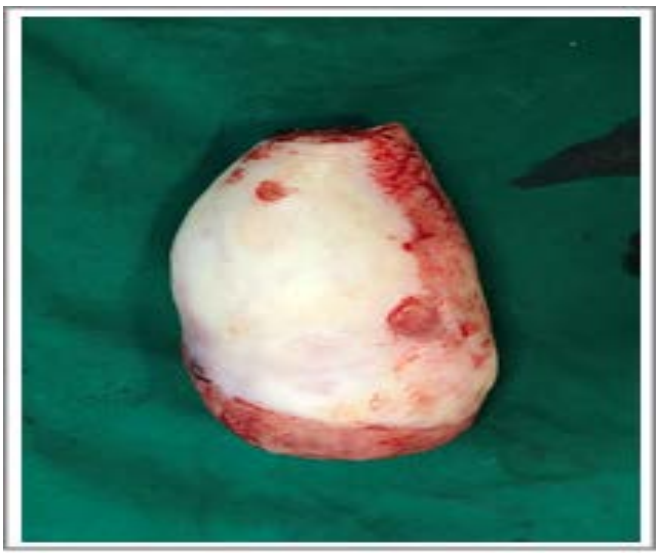

Figure 2 (A): Pink globular mass with ulcerative lake.

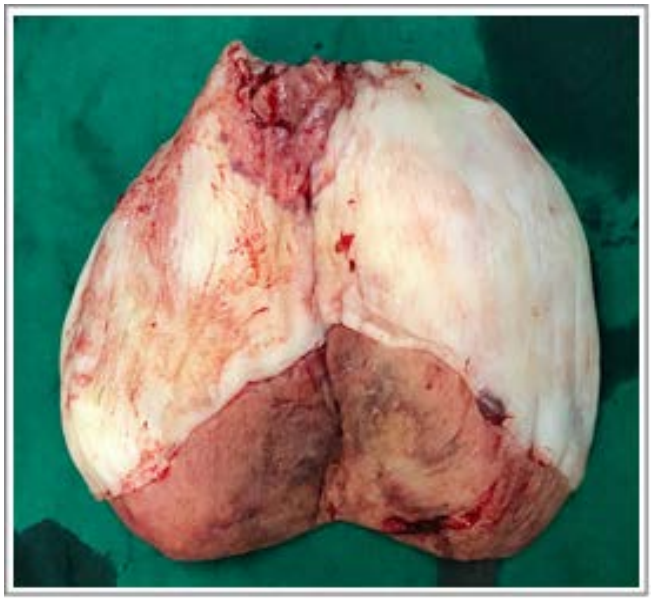

Figure 2 (B): Cut surface is homogenous white with haemorrahgic area.

On microscopic examination plump spindle cells along with many thin and thick blood vessels. Foci of adipocytes and smooth muscle fibers along with a few haemorrhagic spots were seen (Figure 3).

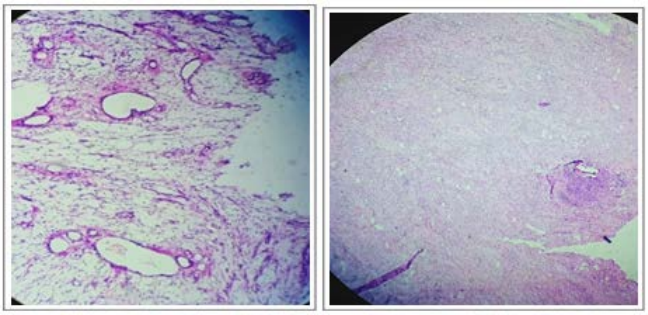

Figure 3: Foci of adipocytes and smooth muscle fibers. Tumor consisting of spindle cells along with blood vessel.

Discussion

Intrauterine Angiomyolipoma of genital tract is a rare tumor, and has not been officially listed in WHO Classification of female reproductive system tumors. It is a benign neoplasm composed of vascular cells, smooth muscle cells and adipose tissue and it belongs to family of Perivascular epithelioid cell tumor (PEComa).[5,6]

Angiomyolipoma are usually seen in association with Tuberous Sclerosis. Other features of Tuberous sclerosis include widespread hamartomas of different organs such as Kidney (angiomyolipoma or renal cyst), Heart (rhabdomyomas), Brain (cortical tubers), bone (sclerotic lesions), eyes (phakomas), lung (lymphangiomyomas), skin (angiofibromas or adenoma sebaceum) and liver. Extra renal angiomyolipomas are rare liver, heart, mediastinum, large bowel, retroperitoneal, nasal cavity, oral cavity, soft tissue, skin, spermatic cord, vaginal wall and fallopian tube. Clinical presentation of angionyolipoma is not specific may present as pain in abdomen, mass coming out of vagina or menorrhagia. Radiologically also the finding depends on gross tumor appearance, not giving significant enough to make an pre-operative diagnosis of tumor. Therefore diagnosis of angiomyolipoma is best made microscopically.[1,7]

Most angiomyolipoma show positive immunoreactivity for HMB-45 and S-100. HMB-45 is positive for the nonvascular smooth muscle cells of Angimyolipomas of Renal and Extrarenal origin, but it has not yet a definitive marker for Cervical or Uterine angiomyolipomas.[8]

Extra renal and Renal Angiomyolipoma have similar histological features, with few differentiating features. Extra renal Angiomyolipoma occurs in older age group, is a well circumscribed lesion that can be easily resected and is HMB-45 negative. While Renal Angiomyolipoma can grow much larger in size, are invasive in nature with tendency to recur. Renal Angiomyolipoma associated with Tuberous sclerosis are positive for HMB-45. Lymphocytic aggregates are associated with extrarenal angiomyolipoma, which is uncommon in renal angiomyolipoma.[9,10]

Few lesion that mimic Angiomyolipoma are Lipoleiomyoma, degenerated myoma, benign lipomatous tumor and vascular leiomyoma with fat component. Lipoleiomyomas show presence of mature adipocytes and muscle cell but vascular component which is a prominent feature of angiomyolipoma. Degenerated myomas can be differeniated from angiomyolipoma by echogenicity, without shadowing and irregular margins.[8,11,12]

Angiomyolipoma are usually benign, however they may be associated with haemorrhage and invasion of surrounding organs. Therefore, recently angiomyolpioma is considered as a slow growing malignant neoplasm with ability to metastasize.[13,14]

Conclusion

Angiomyolipoma although a rare entity should be considered as differential diagnosis for Abdominal mass and dysfunctional uterine bleeding. Research for the better understanding of the nature of the disease, histogenesis, malignant potential and management of the disease is needed.

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Abbreviations

Angiomyolipoma (AML)

Perivascular epithelioid cells tumors (PE Coma)

Clear cell “sugar” tumor of lungs (CCST)

Lymphangioleiomyomatosis (LAM)