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ANGIOMYOLIPOMATOUS HAMARTOMA - A CASE REPORT WITH REVIEW OF LITERATURE

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ABSTRACT

Angiomyolipomatous hamartoma is a rare benign vascular disease. The tumor is characterised by replacement of parenchymal lymphoid tissue by blood vessels, adipose tissue, smooth muscle and fibrous tissue. The most common lymph node involved are inguinal and femoral lymph nodes. Angiomyolipomatous hamartoma has a wide range of age distribution ranging from 8 months to 89 years with male predominance. The common clinical presentations are painless, non-tender and mobile mass. Angiomyolipomatous hamartoma is important to recognise because it clinically mimic various tumors. Differential diagnosis to be considered are lymphangiomatosis, leiomyomatosis and angiomyolipoma of lymph node. We described angiomyolipomatous hamartoma keeping in mind its unusual occurance and in order to elucidate its biological potential and histogenesis.

Key words: Angiomyolipomatous Hamartoma, Benign Vascular Tumor, Inguinal Lymph Node.

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INTRODUCTION

Case report

A 54 year old female presented with the complaints of swelling in the right inguinal region for past one year. On physical examination a mobile, non-tender and firm swelling measuring 2x1cm was identified in the right inguinal region. All the laboratory investigations were within normal range. Radiological examination showed features of benign soft tissue tumor, in favour of lipoma. The mass was resected and sent for histopathological examination. Post-operative period was uneventful. Grossly we received a single fibro-fatty tissue mass measuring 2x1x1cm. External surface grey yellow and greasy, cut surface grey yellow to grey white, greasy and soft in consistency. Microscopic examination revealed a lymph node replaced by mature adipocytes, smooth muscle, fibrous tissue and numerous blood vessels of varying size. These features were prominently extending from hilum to the convex surface of lymph node. These histomorphological features were in favour of angiomyolipomatous hamartoma.

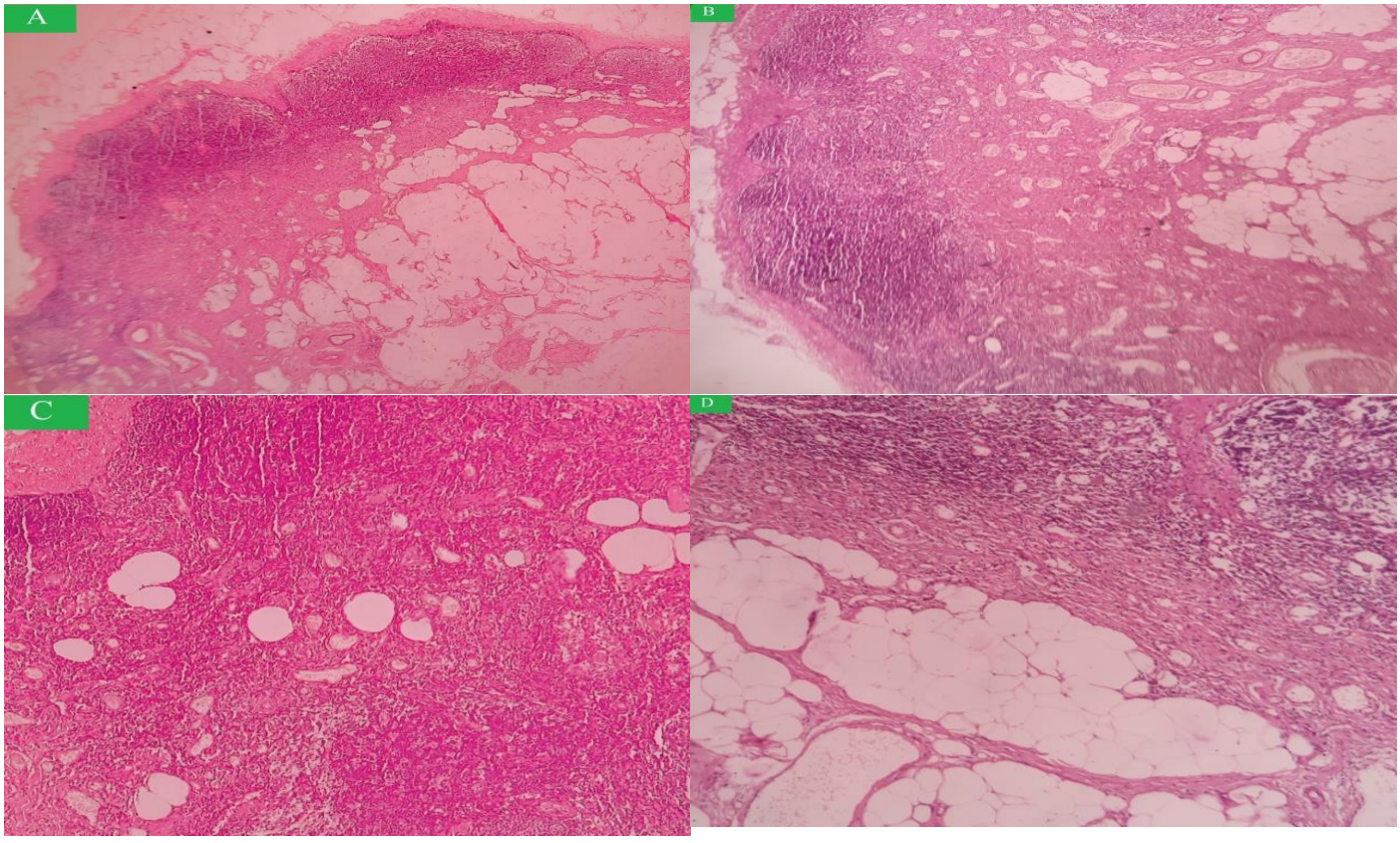
DISCUSSION

Angiomyolipomatous hamartoma is a rare benign vascular disease of unknown etiology.[1] This entity was first described by Chan et al in 1922. The tumor is characterised by replacement of parenchymal lymphoid tissue by blood vessels, adipose tissue, smooth muscle and fibrous tissue.[2] The most common lymph node involved are inguinal and femoral lymph nodes. However several cases have been reported in cervical and popliteal lymph nodes.[3-5] In the present study, we report a case of angiomyolipomatous hamartoma in the inguinal lymph node. Among the reported cases it is observed that the male to female ratio is about 1.6:1.[6,7] The tumor has a wide range of age distribution ranging from 8 months to 89 years.[8] In general most of the patients are asymptomatic,

but few patients present with symptoms of non-tender and mobile mass.[9] In the present study 54 years female patient presented with the complaints of swelling in the right inguinal region for past one year. Gross presentation of angiomyolipomatous hamartoma is unencapsulated single or multiple yellow, fatty nodules of varying size and soft in consistency.[6] In the present study, grossly we received a single fibro-fatty tissue mass measuring 2x1x1cm. External surface and cut surface grey yellow to grey white, greasy and soft in consistency. The histomorphology of angiomyolipomatous hamartoma shows lymphoid tissue completely replaced with lobules of mature adipocytes, bundles of smooth muscle cells and fibrous tissue in a disorganized fashion. In some cases few atrophic lymphoid follicles in the subcapsular area were identified. The microscopic features observed in the present study revealed a lymph node replaced by mature adipocytes, smooth muscle, fibrous tissue and numerous blood vessels of varying size. Also few atrophic lymphoid follicles were maintained.

Angiomyolipomatous hamartoma is important to recognise, because it clinically mimic various tumors. Differential diagnosis to be considered are lymphangiomatosis, leiomyomatosis and angiomyolipoma of lymphnode. The histomorphology of lymphangiomatosis is characterized by the presence of smooth muscle cells arranged in fascicles and sheets around anastomosing vascular spaces forming a pericytomatous pattern.[10] The microscopic feature of Angiomyolipoma of the lymphnode shows haphazardly arranged adipose tissue, smooth muscle cells and thick walled blood vessels. Smooth muscle may be epithelioid or pleomorphic with infrequent mitoses and the tumor show positivity for melanoma associated antigen HMB-45.[11] The microscopic feature of Nodal leiomyomatosis is composed of smooth muscle cells proliferation in compact bundles with no vascular component and studies have been reported, it has an association with HIV and immunocompromised patients.[12] Role of Immunohistochemistry in Angiomyolipomatous hamartoma are Alpha smooth muscle actin (ASMA) and Desmin shows strong positivity for smooth muscle cells. CD 34 shows positivity for blood vessels.[13] Recurrence and metastasis of angiomyolipomatous hamartoma has not been reported, hence extensive resection is not indicated.

[Fig-1]: (A & B) Microphotography showing a thin rim of residual lymphoid tissue replaced by smooth muscle cells, blood vessels, fibrous and adipose tissue(Stain: Haematoxylin and eosin, 10X). (C & D) Microphotography showing lymphoid tissue admixed with mature adipose tissue, smooth muscle, blood vessels and fibrous tissue (Stain: Haematoxylin and eosin, 40X)



CONCLUSION

Angiomyolipomatous hamartoma is a rare benign vascular tumor. The tumor is characterised by replacement of parenchymal lymphoid tissue by blood vessels, adipose tissue, smooth muscle and fibrous tissue. We described angiomyolipomatous hamartoma keeping in mind its unusual occurrence and in order to elucidate its biological potential and histogenesis.

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