International Surgery Journal
Ravikumar G et al. Int Surg J. 2016 Aug;3(3):1691-1696
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: http://dx.doi.org/10.18203/2349-2902.isj20162779

Case Report

Myoid hamartoma breast with pseudo angiomatous stromal hyperplasia: a case report and review of literature

Ravikumar G.[1]*, Sivakami T.[2], Sugapradha GR[3]

[1]Department of Plastic Surgery, Thanjavur Medical College, Thanjavur, Tamilnadu, India
[2]Department of Anatomy, Thanjavur Medical College, Thanjavur, Tamilnadu, India
[3]Multidisciplinary Research Unit, Thanjavur Medical College, Thanjavur, Tamilnadu, India

Received: 13 June 2016
Accepted: 15 July 2016

***Correspondence:**
Dr. Ravikumar G.,
E-mail: gravikumar1962@gmail.com

ABSTRACT

Hamartomas of the breast, also known as fibroadenolipomas, lipofibroadenomas or adenolipomas are benign lesions with low risk of recurrences. Hamartomas account for between 0.04 and 1.15% of all benign breast tumours in females. Myoid hamartoma of the breast (MHB) with Pseudo Angiomatous Stromal Hyperplasia (PASH) is an extremely rare lesion. These cases will be misdiagnosed preoperatively. Histopathology only gives definitive diagnosis. Sixteen year old girl admitted with a mass of two months duration in the left breast in which reduction mammoplasty was done two years back. Ultra sonogram of the left breast revealed a well circumscribed 10cm x 6 cm x 3cm mass with echogenic and isoechoic areas. A core needle biopsy was done and reported as fibro adenoma of the left breast. Excision of the mass with adequate margins was done. The histopathology report of the resected mass showed myoid hamartoma breast with pseudo angiomatous stromal hyperplasia (PASH). So far there is no literature report about myoid hamartoma breast with pseudo angiomatous stromal hyperplasia (PASH) in an already reduction mammoplasty done patient. We present this case for its rarity and also to stress the importance of immunohistochemical studies in benign breast lesions.

Keywords: Myoid hamartoma, Breast, PASH

INTRODUCTION

Hamartomas of the breast, also known as fibroadenolipomas, lipofibroadenomas or adenolipomas, are benign lesions that were first described in 1971.[1] Hamartomas account for between 0.04 and 1.15% of all benign breast tumours in females.[2] Myoid hamartoma of the breast is composed of differentiated mammary glandular and stromal structures and is considered to be a rare variant of mammary hamartoma. Myoid hamartomas may present as painless breast lumps and hyper echoic or mixed echogenic on ultrasound examination. Myoid Hamartoma of the breast with Pseudoangiomatous stromal hyperplasia (PASH) is very rare.

CASE REPORT

A sixteen year old female had undergone reduction mammoplasty on the right breast two years ago in other hospital. (Figure 1, 1a) Three months later reduction mammoplasty of the left breast was done in our hospital. (Figure 2, 2a, 2b) The histopathology report of the reduction mammoplasty specimen was diffuse hyperplasia of the breast.

Now she got admitted with a mass of two months duration in the left breast. She noticed the swelling on self-examination and it was increased gradually. There was no history of nipple discharge or pain in the left breast. Clinical examination of the left breast revealed a mobile, firm, lobulated mass of 10cm x 6cm in the upper outer quadrant. (Figure 3) Right breast and both axillae

were normal. Ultra sonogram of the left breast revealed a well circumscribed mass with echogenic and isoechoic areas. A core needle biopsy was done and reported as fibro adenoma of the left breast. Excision of the mass with adequate margins was done (Figure 3a).

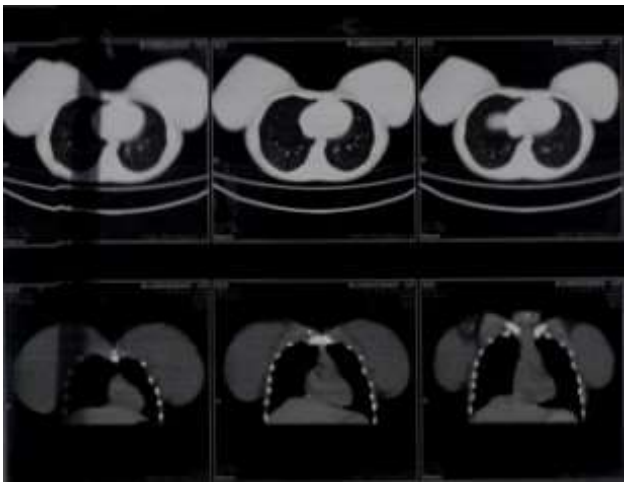

Figure 1: MRI showing bilateral giant hypertrophy of breasts.

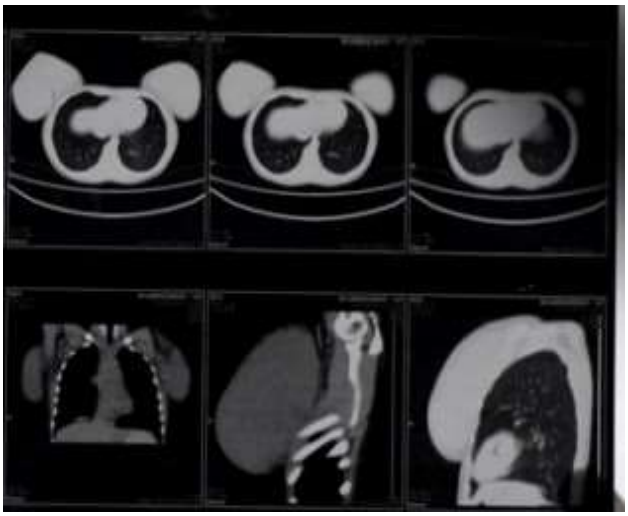

Figure 1a: MRI showing giant hypertrophy of breasts -side view.

Gross examination of specimen showed a 12cm x 12cm x 7cm soft tissue mass, nodular, greyish white on cut section with tumour free resected margins. (Figure 3b, 3c) Histopathology showed a well encapsulated tumour composed of breast lobules and ductules formed by ductal and myoepithelial cells (Figure 4). The lobules were surrounded by spindle cell stroma. In many areas the stroma was hyalinised with pseudoangiomatous spaces (Figure 4a). No nuclear atypia could be made out. Immuno histochemical study revealed highly positive progesterone receptors in ductal epithelial cells (Figure 4b). Highly positive CD34 in stromal cells, (Figure 4c) positive Actin in stromal spindle cells, (Figure 4d) and positive BCl2 in the stromal nuclei (Figure 4e) and negative CD31 in pseudo vascular areas (Figure 4f). Consequently a diagnosis of Myoid Hamartoma breast with Pseudoangiomatous stromal hyperplasia (PASH) was made. Six months follow up of the patient showed no recurrence of the mass.

Figure 2: Reduction mammoplasty of right breast was done already. Reduction mammoplasty of left breast was planned-preoperative markings shown.

We present this case for the rare diagnosis, large size of the tumour, occurrence of the tumour on a reduction mammoplasty done breast which is so far not reported in the literature, the role of immune histochemical study in benign breast lesions and to stress the significance of follow up as hamartoma has the potential for malignant transformation.

DISCUSSION

Arrigoni first coined the term breast hamartoma in 1971 although it had been described earlier under various terms since 1904 by Albrecht.[1,2] Breast hamartoma is a poorly recognised benign breast lesion, its incidence varies between 0.04 and 1.15%.[2] Breast hamartomas also known as fibroadenolipomas, lipofibroadenomas or adenolipomas, encompass a collective overgrowth of mature tissues lacking in organisation. The tissues include benign ductal and lobular units, adipose tissue, fibrous stroma and smooth muscle tissue. Hamartoma with a marked component of smooth muscle tissue is known as myoid hamartoma. This term was first used by Davies and Riddell in 1973.[3] Myoid hamartomas are very rare variants of breast hamartoma. According to the literature the average age of the patients with breast

hamartoma ranged between 19-56 years. [4] The patient's age in our case was 17 years which is the lowest age reported so far in the literature.

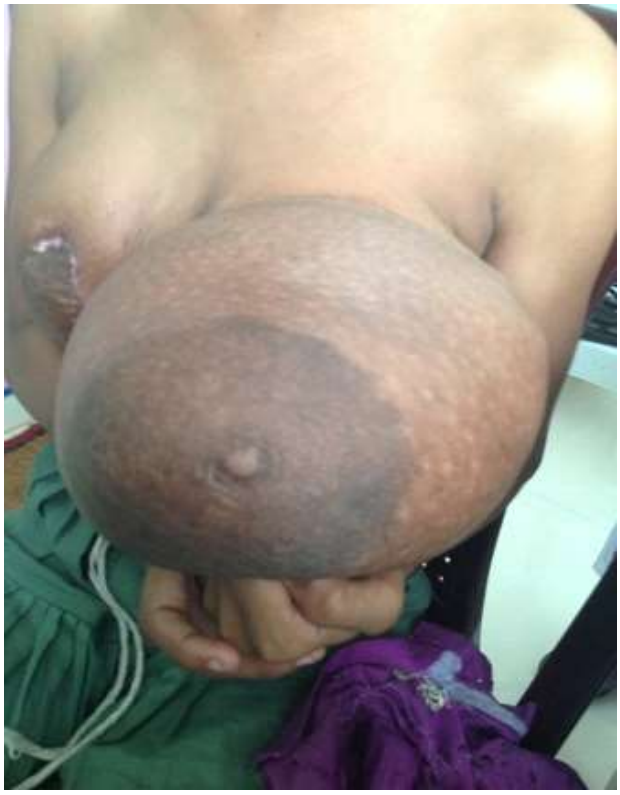

Figure 2a: Hypertrophied pendulous left breast.

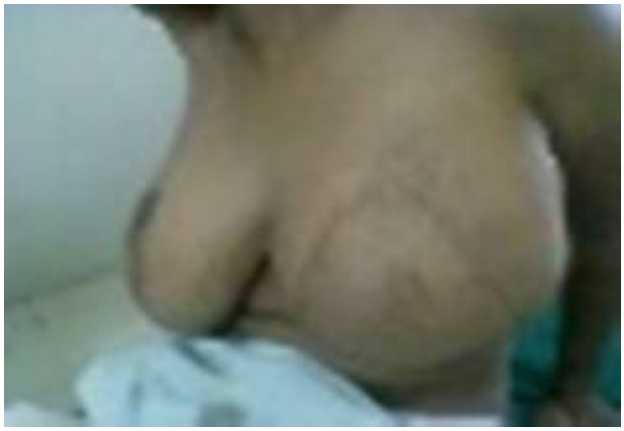

Figure 2b: One year after bilateral reduction mammoplasty.

Clinically, myoid hamartomas present as firm, mobile, well-defined, non-tender breast lumps which may be mistaken as fibro adenoma. Mammography usually reveals a sharply delineated dense mass, occasionally with calcifications, none of which are usually suspicious. Ultra-sonographic examination normally confirms the solid nature of the lesion. This tumor generally has an admixture of smooth muscle stroma with entrapped ductules, and lobules and foci of mature adipose tissue. The possible sources of smooth muscle proliferation in mammary tissue are blood vessel walls, myoepithelium, undifferentiated mammary mesenchyme and muscularis mammillae of areolae [5]

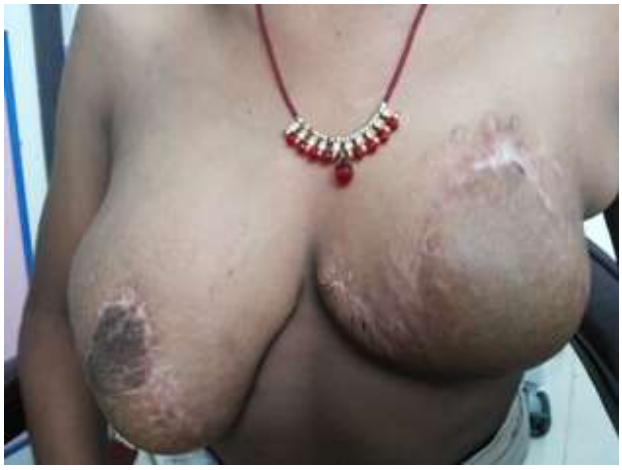

Figure 3: Recurrent mass in the upper outer quadrant of the left breast.

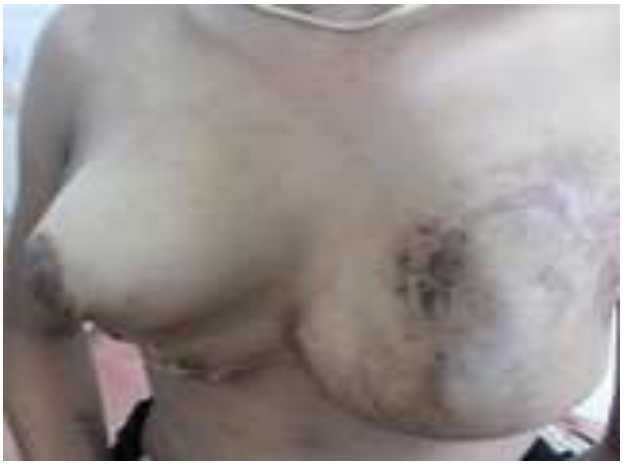

Figure 3a: After excision of mass in left breast and reduction mammoplasty of right breast.

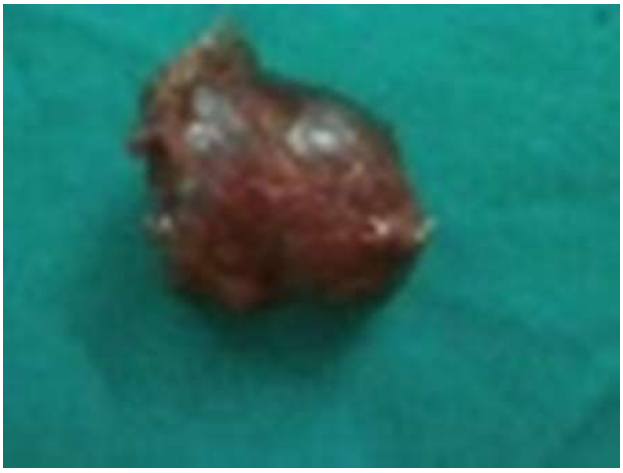

Figure 3b: Gross examination of the myoid hamartoma of the breast.

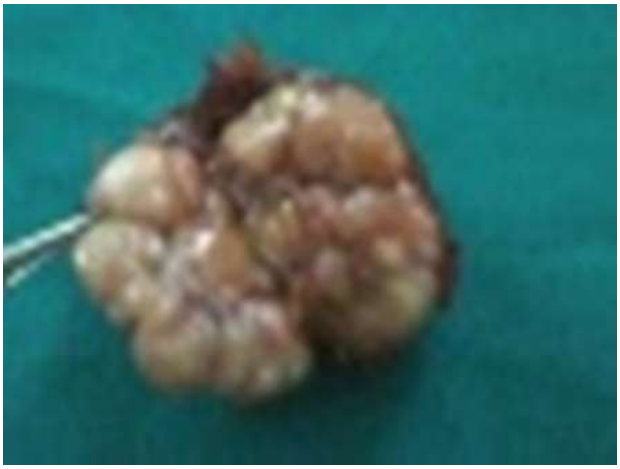

Figure 3c: Cut section of the myoid hamartoma of the breast.

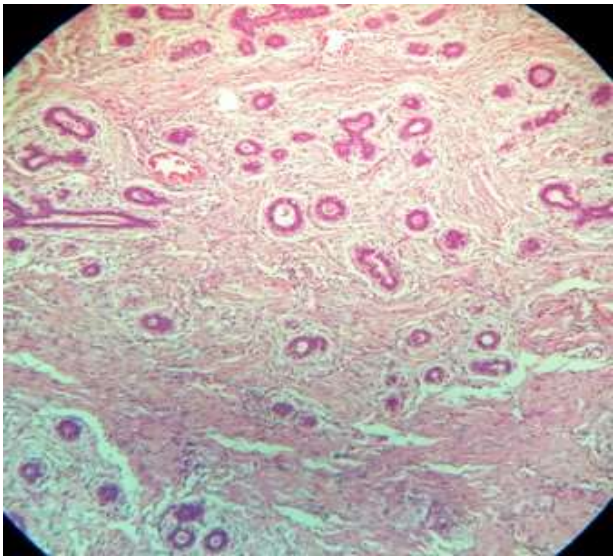

Figure 4: Well encapsulated tumour composed of breast lobules and ductules formed by ductal and myoepithelial cells.

Various theories had been proposed to explain the presence of this heterologous component of smooth muscle. Amongst the theories is the differentiation from a common stromal cell to evolve into smooth muscle cells.[6] The expression of CD34 in smooth muscle cells in myoid hamartoma helps support this origin from a common stromal cell. This also explains the presence of a variety of heterologous components viz. cartilage and bone found in breast hamartomas and other breast stromal tumours. [7]

Fine needle or core needle biopsies may not reveal the diagnosis but contain benign breast components which may be reported as fibroadenoma. Cytology of myoid hamartoma is also non-specific and overlaps with other benign breast lesions. Herbert et al reviewed eight cases of breast hamartoma and found none of them had a preoperative FNA diagnosis of hamartoma.[8]

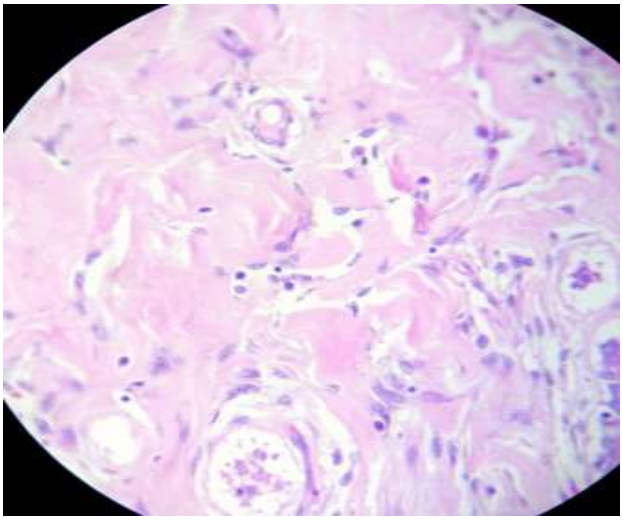

Figure 4a: The lobules were surrounded by spindle cell stroma. In many areas the stroma was hyalinised with pseudoangiomatous spaces.

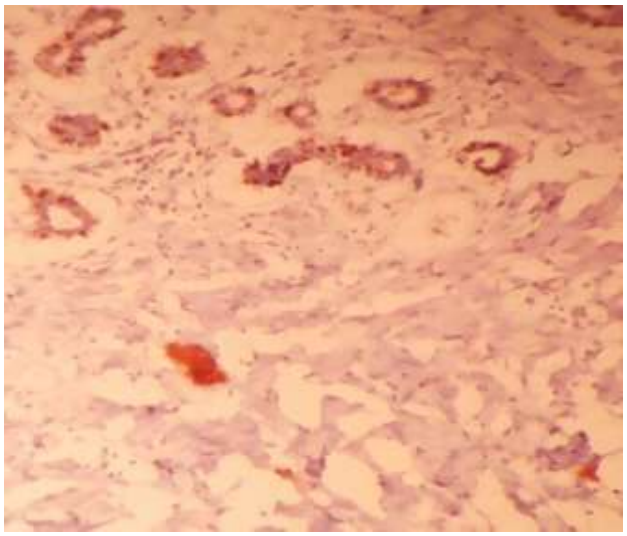

Figure 4b: Immuno histochemical study showing highly positive progesterone receptors in ductal epithelial cells.

Various differential diagnoses for myoid hamartoma include fibro adenoma with smooth muscle metaplasia, leiomyoma, benign nerve sheath tumour, pleomorphic adenoma of breast and fibromatosis. Other malignant breast lesions that need to be considered include mesenchymal metaplasia carcinomas with chondroid and osseous components.

Definitive diagnosis of myoid hamartoma is made by histology and immunohistochemistry. Myoid hamartoma is differentiated from fibro adenoma by the lack of an intracanalicular or pericanalicular pattern of ductal and stroma proliferation. It also differs from leiomyoma by containing entrapped normal ductal and lobular units amongst the smooth muscle bundles, not seen in leiomyoma.

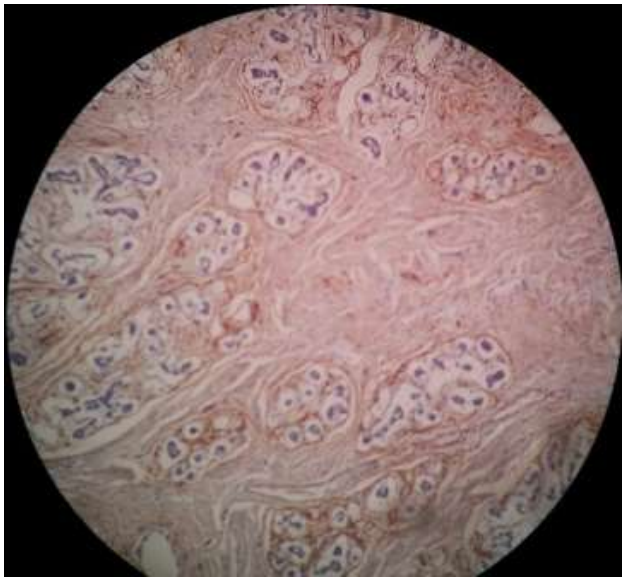

Figure 4c: Immuno histochemical study showing highly positive CD34 in stromal cells.

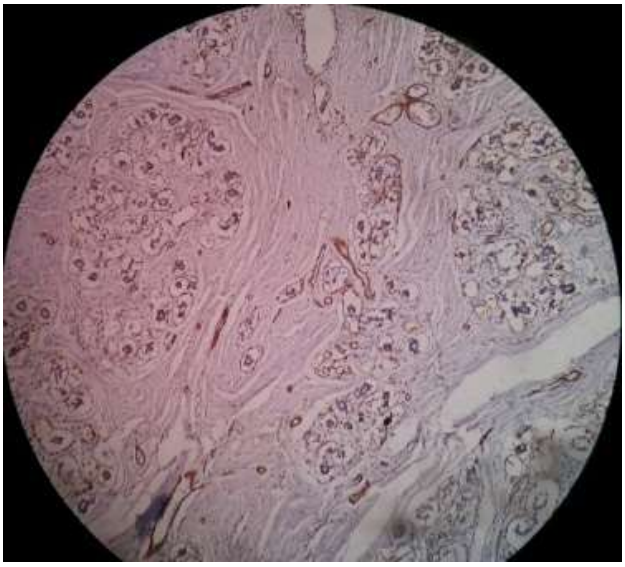

Figure 4d: Immuno histochemical study showing positive actin in stromal spindle cells.

Immunohistochemical studies are helpful in making a diagnosis, and several reports have suggested that spindle and epithelioid tumor cells show strong positive staining for SMA, desmin, and vimentin, and the absence of staining for cytokeratin as well as S-100 protein.[9]

Our case revealed highly positive progesterone receptors in ductal epithelial cells, highly positive CD34 in stromal cells, positive Actin in stromal spindle cells, positive BCl2in the stromal nuclei and negative CD31 in pseudo vascular areas. Myoid hamartomas are benign breast lesions with low risk of recurrences.[10] It is also very rare but coincidental malignancies have been reported.[10] Thus, wide excision of the lesion with detailed histopathological examination is recommended treatment for myoid hamartomas.

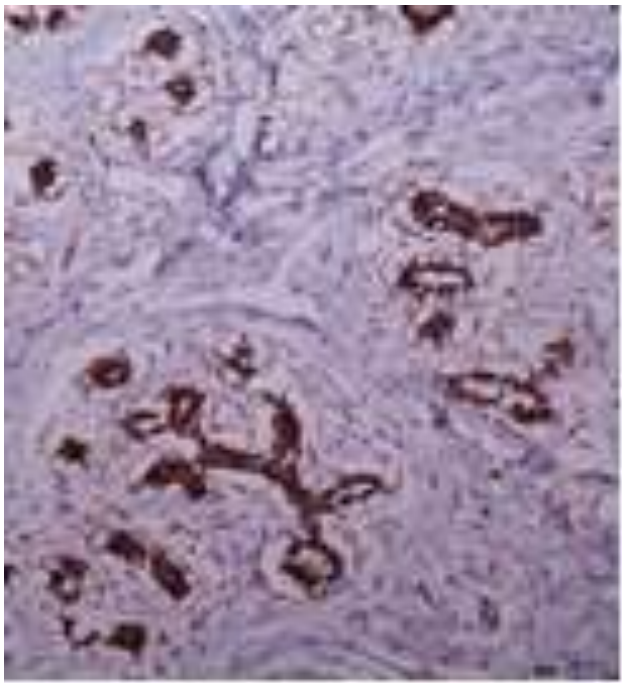

Figure 4e: Immuno histochemical study showing positive BCL2 in the stromal nuclei.

Figure 4f: Immuno histochemical study revealed negative CD31 in pseudo vascular areas.

CONCLUSION

Myoid hamartoma breast with pseudo angiomatous stromal hyperplasia (PASH) is very rare. Mammography, ultrasonography or fine needle aspiration cytology may not give the definitive diagnosis. Breast hamartomas are usually diagnosed as fibro adenomas. Immunohistochemical studies must be done in all benign breast lesions as hamartoma has got the potential for malignant transformation. Adequate tumour free margin during resection and follow up are essential for all benign breast lesions.

ACKNOWLEDGEMENTS

Authors would like to thanks to the Department of Plastic Surgery, Thanjavur Medical College.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Ravikumar G., Sivakami T., Sugapradha GR. Myoid hamartoma breast with pseudo angiomatous stromal hyperplasia: a case report and review of literature. Int Surg J 2016;3:1691-6.