Mammary hamartoma- a clinical dilemma

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Abstract

Mammary hamartoma is an uncommon benign lesion. The frequency of the tumor is reported between 0.04 -1.15% and it accounts for 4.8% of all benign breast tumors. The lesion may be under-diagnosed by the clinician because of the varied clinical presentation or by the pathologist, as the microscopic appearance is not distinctive. A case of left sided mammary hamartoma in a 46-year-old female who presented with a freely mobile mass is reported. A clinical diagnosis of fibroadenoma was made and lumpectomy performed. Histopathology and gross diagnosis confirmed it to be a case of mammary hamartoma. Extensive area of adenosis, focal cystic change, apocrine metaplasia and columnar cell change was noted. Identification of hamartoma is important because there are the problems of recurrence, co-existent carcinomas and association with Cowden syndrome.

Keywords:

Hamartoma, fibroadenoma, apocrine metaplasia, ductal carcinomas

Introduction

Hamartomas are relatively uncommon lesions which occur predominantly in the peri-menopausal age group [1-4]. The term hamartoma was first coined by Arrigoni et al in 1971, as a well-circumscribed breast lesion with varying amounts of benign epithelial elements, fibrous tissue and fat [1]. Many authors consider this entity to be under-diagnosed [3,4]. Clinically, they may be confused with both fibroadenoma and phyllodes tumor. Radiologically, they are large, well-circumscribed mass lesions with central lucency. Although they may cause marked distortion of the breast, hamartomas are entirely benign. They may present as a large palpable mass but surprisingly, they are often impalpable and detectable only mammographically.

Case Report

A 46-year-old lady visited the department of surgery with a complaint of recurrent painless lump in her left breast, with no nipple discharge, which was noticed 2 years ago. She had noticed a recent increase in size of the swelling. Examination revealed a palpable freely mobile soft mass measuring 5x4 cm and there was no axillary lymphadenopathy. There was a previous history of bilateral breast swellings in both breasts 10 years back which was surgically removed and the histopathology diagnosis made was bilateral fibroadenomas. There was no family history of breast lesions and malignancies. The previous histopathology slides with the diagnosis of bilateral fibroadenoma could not be retrieved for slide review. The patient did not have manifestations of Cowden syndrome with which it may be associated.

Laboratory values such as Hb, Total count, Differential count, ESR, Blood sugar, Urine routine examination were all within normal limits. Since the patient insisted on excision of the lesion and refused prior FNAC and imaging studies, a preoperative clinical diagnosis of fibroadenoma was made and a lumpectomy was performed. Preoperatively even though a freely mobile mass was felt, intraoperatively it was soft and impalpable. Excision biopsy was performed with a circumareolar incision. There was difficulty in extracting the lesion at focal area. The post-operative period was uneventful and the patient was discharged on the third post-operative day.

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Fig. 1: Well-circumscribed lesion with adipose tissue in the center (arrow pointing to adipose tissue)

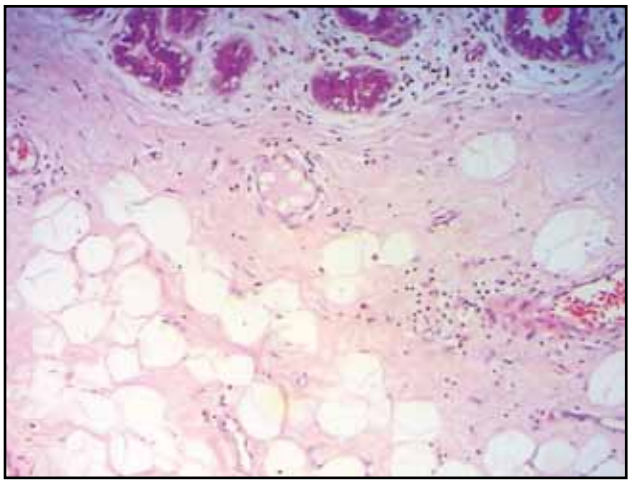

Fig. 2: Adipose tissue between breast lobules (H & E, x 10)

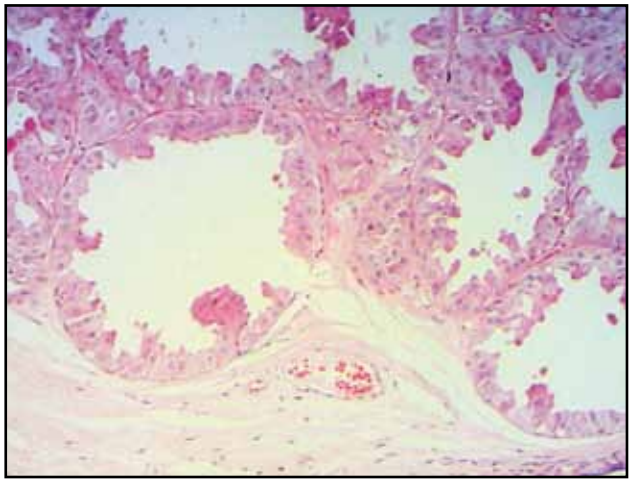

Fig. 3: Areas with apocrine metaplasia (H & E, x 40)

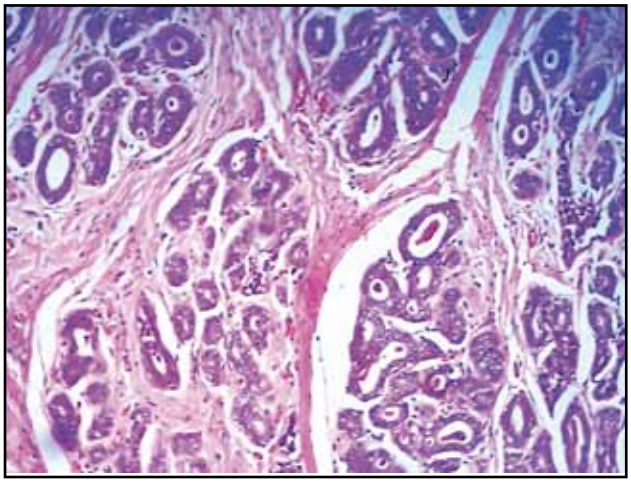

Fig. 4: Areas with extensive adenosis (H & E, x 10)

A well-circumscribed ovoid mass measuring 45x40mm with firm to soft consistency was received in the pathology department. Cut surface of the mass showed fat in the center of the lesion which raised the suspicion of hamartoma (Fig. 1). A few tiny cysts were also detected grossly. The nodular structure and slit like spaces characteristic of a fibroadenoma was not seen. Histopathological examination confirmed the diagnosis of mammary hamartoma which showed adipose tissue predominantly in the center of the lesion between breast lobules (Fig.2). The stroma was fibrotic and not fibromyxoid as in fibroadenoma. Focal cystic change, apocrine metaplasia (Fig. 3), extensive areas of adenosis (Fig 4), and columnar cell change were also noted.

No problem was detected during follow-up for 3 months. However, an alert for follow up was advised since there are chances of recurrence and malignant transformation in mammary hamartomas.

Discussion

Mammary hamartomas are breast disorders currently underestimated and not well recognized. Hamartomas accounted for 1.2% of benign lesions [5] and 4.8% of benign breast tumors [6]. The true incidence is probably higher as pointed out by Daya et al [4] and substantiated by Tse et al [7]. Hamartomas result more from breast dysgenesis than from tumorous process.

Clinically, they may be confused with both fibroadenoma and phyllodes tumor. The lesions vary considerably in size from 1 to 15 cm, although the majority measure between 2 and 5 cm. They form well-circumscribed oval masses which have a firm fleshy consistency. Microscopically, mammary hamartomas lack a true capsule, although they separate easily from

adjacent breast tissue. They are composed of variable mixtures of connective tissue stroma and breast lobules. The latter usually have a normal configuration. Occasional ectatic ducts are present and these may become cystic. Epithelial hyperplasia is not a feature. Islands of adipose tissue are found frequently, often in the center of the lesion. The presence of pseudo-angiomatous stroma within the hamartoma has been reviewed and described in detail by Fisher et al [3]. Studies done by Tse et al [7] in 25 patients showed adipose tissue in all cases and interlobular fibrosis in 21 cases. Benign epithelial hyperplasia occurred in 10 cases, and pseudo-angiomatous stromal hyperplasia or cystic ducts in eight cases each. Apocrine metaplasia, calcification, stromal giant cells, and adenosis occurred in four cases or less. In our patient we were able to identify extensive area of adenosis which is rarely reported. Two cases showed coexisting ductal carcinoma *in situ* limited to within the hamartoma.

Imaging modalities can aid in the diagnosis of mammary hamartoma. Imaging (mammography, ultrasound and magnetic resonance imaging) was performed in 18 cases of mammary hamartoma showed encapsulated masses with a heterogeneous appearance [7]. However, the degree of mammographic opacity is variable and depends upon the fat /parenchyma ratio[5]. Breast hamartoma has a wide variation of sonographic appearance, which is not helpful for differential diagnosis [8]. FNAC may also help in preoperative diagnosis of mammary hamartoma. The conclusion of studies done by Herbert et al[9] showed that the finding of intact lobular units and a relative paucity of stroma may suggest the diagnosis of hamartoma.

The clinical dilemma in this case was the palpable freely mobile soft breast swelling which led to a diagnosis of fibroadenoma. Intraoperatively the lesion was soft and difficult to locate which was against the findings of fibroadenoma which are palpable lesions. The third dilemma was that usually fibroadenoma and mammary hamartoma can be enucleated but here there was difficulty in focal areas in extracting the lesion.

On histopathology examination there was the presence of extensive area of adenosis. There have been reports of adenosis but the presence of extensive area of adenosis is rarely reported.

Mammary hamartomas are usually soft to firm and can be shelled out at the time of surgery. Here a focal area was difficult to enucleate which could have been due to fibrosis induced by previous surgery. Although no tumor recurrence was observed in most series [3, 6] breast hamartomas occasionally recur after local excision [4]. The previous excision biopsy done bilaterally 10 years ago may have been mammary hamartoma (slides were not available) and hence mammary hamartoma recurrence may be a possibility.

Mammary hamartomas can coexist with ductal carcinoma *in situ* [10]. Lobular carcinomas [11, 12] and ductal carcinomas were found associated with mammary hamartoma [13]. Mester et al[14] reported a case with *in situ* and infiltrating ductal carcinoma arising in a breast hamartoma. But when associated with Cowden's syndrome, mammary hamartomas are diffuse or bilateral lesions [15]. This patient did not have features of Cowden syndrome.

We may conclude that the soft mobile mass in the breast impalpable intraoperatively could be mammary hamartoma. The diagnosis may be aided by imaging studies and FNAC. A keen awareness of the lesion by the pathologist finally helps in diagnosis even if the above modalities are not available. The importance of diagnosing mammary hamartoma is due to the possibility of its association with malignancy, recurrence and Cowden syndrome.

References

1. Arrigoni M G, Dockerty M B, Judd E S, et al. The identification and treatment of mammary hamartoma. Surg Gynec Obstet 1971;132:259–262
2. Linell F, Ostberg G, Soderstrom J, et al. Breast hamartomas. An important entity in mammary pathology. Virchow's Arch (A) Anat Pathol 1979;383:253–264
3. Fisher CJ, Hanby AM, Robinson L, et al. Mammary hamartoma—review of 35 cases. Histopathol 1992;20:99–106.
4. Daya D, Trus T, D'Souza TJ, et al. Hamartoma of the breast, an underrecognized breast lesion. Am J Clin Pathol 1995;103:685–9.
5. Hessler C, Schnyder O, Ozello L, et al. Hamartoma of the breast: diagostic observation of 16 cases. Radiol 1978;126:95-8.
6. Charpin C, Mathoulin MP, Andrac L, et al. Reappraisal of breast hamartomas: a morphological study of 41 cases. Pathol Res Pract 1994; 190:362–371.
7. Tse GM, Law BK, Ma TK, et al. Hamartoma of the breast: a clinicopathological review. J Clin Pathol 2002;55:951–954.
8. Adler DD, Jeffries DO, Helviem A, et al. Sonographic features of breast hamartomas. Ultrasound Med 1990,9: 85-90.
9. Herbert M, Mendlovic S, Liokumovich P, et al. Can hamartoma of the breast be distinguished from fibroadenoma using fine-needle aspiration cytology? Diagn Cytopathol 2006;34:326-9.
10. Tse GM, Law BK, Pang LM, et al. Ductal carcinoma in situ arising in mammary hamartomas. J Clin Pathol 2002;55:541–2.
11. Coyne J, Hobbs FM, Boggis C, et al. Lobular carcinoma in a mammary hamartoma. J Clin Pathol 1992;45:936–7.
12. Kuroda N, Sugimoto T, Numoto S, et al. Microinvasive lobular carcinoma associated with intraductal spread arising in a mammary hamartoma. J Clin Pathol 2002;55:76–77.
13. Anani PA, Hessler CH. Breast hamartoma with invasive ductal carcinoma. Report of two cases and review of the literature. Pathol Res Pract 1996;192:1187–94.
14. Mester J, Simmons RM, Vazquez MF, et al. In situ and infiltrating ductal carcinoma arising in a breast hamartoma. Am J Roentgenol 2000;175:64–6.
15. Pawar VR, Shinde UV, Patil PP, et al. Breast Hamartoma. Indian Medical Gazette — January 2013; 37-38