

Case Report- A case of Primary Bilateral Fibroadenoma of Breasts in A male of 20 years of age.

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Abstract- We are reporting here a case of a healthy male age 20 years, presented with bilateral gynaecomastia with hard mobile swelling in each breast. Fnc showed normal breast tissue & benign fibrocystic disease in the swelling of each breast. Bilateral simple mastectomy done. Specimen hpe showed fibroadenoma in normal breast tissue on both sides. This case is being reported for it's unique presentation.

HISTORY-

A 20 years healthy male presented with bilateral gynaecomastia. The breasts started enlarging from the age of 14 years of age & grown to the present size. No history of pain / rapid increase in size of the breasts. He noticed a swelling in each of the breasts since last 6 months gradually increasing in size & occasionally painful if squeezed forcefully or traumatized. History of Development of secondary sexual characters as a male, was normal i.e. Growth of penis, testes, facial & pubic hair, hair on extremities, muscle developments, ejaculations etc.etc. No history of similar case in family / chromosomal aberrations in family. Not a product of consanguineous marriage.

CLINICAL EXAMINATION-

Wel built & nourished fair skinned male having bilateral fully grown breasts equivalent to a female of same age, having a hard mobile lump of a size of 4c.m. X 3.5 c.m. In right breast & a lump of a size of 5c.m. X 4 c.m. In left breast, freely mobile within the breast tissue, nontender. Nipple & areola normal. No discharge through the nipple. No palpable / suspicious nodes in axillae.

growth of penis, testes, hair pattern on face, extremities normal as a male. Testicular sensation preent. Cremasteric reflex normal.

INVESTIGATIONS-

Usg- bilateral normal breast tissue with presence of mobile lumps of size 3.5c.m. X 3c.m. On right side & 4.5cm x3.5c.m. On left side within the breasts.

Usg- abdomen & pelvis- no e/o uterus, ovaries or adnexa – [normal size or rudimentary.]

bladder, prostate, seminal vecicles normal.

Chromosomal studies- karyotyping showed normal pattern of 44xy.

Blood investigations- s. Testesteron levels normal for his age but s.

Estrogen higher level of normal limits.

MANAGEMENT-

Simple mastectomy done on both sides under ga. Hpe of both the specimens showed normal breast tissue with fibroadenoma of ----- type. Post op recovery was uneventful. One year follow up- no recurrence of gynecomastia, no lump in breast area, no suspicious nodes in axillae.

Review of literature / discussion-

many cases have been reported of bilateral gynecomastia in males of all ages [primary as well as secondary to liver diseases, ca. Prostate etc.etc.] & bilateral fibroadenomas in elderly males secondary to ca. Prostate after estrogen therapy. However primary bilateral fibroadenomas in both breasts along with gynecomastia in a healthy male who is having 44xy chromosomal pattern is a unique presentation.

Fibroepithelial lesions are uncommon in the male breast. Most published reports describe phyllodes tumors. Fibroadenomas are very common in female breasts, but are exceedingly rare in the male breast. Gynecomastia and/or lobular differentiation have been known to coexist in both types of fibroepithelial lesions in men

We report an exceptional case of recurrent, bilateral fibroadenomas in a man under treatment for prostatic

carcinoma. Although very common in female patients, fibroadenomas are exceedingly rare in the male breast. In fact, some question their existence. For instance, Holleb et al² stated that no true fibroadenoma is formed in men; others have stated that the lesions reported to be fibroadenomas by some authors are poorly documented or appear to be nodular foci of gynecomastia.³ Nonetheless, fibroadenomas in the male breast have been documented sporadically in the medical literature as single case reports and in a rare series of 4 patients.⁴⁻¹² Less convincing reports include a 15-year-old adolescent who presented with a fibroepithelial lesion and concurrent gynecomastia, in which the reported presence of "occasional mitotic figures" in the stroma raises suspicion that this represents a phyllodes tumor and not a fibroadenoma.⁸ In our patient, the phenomenon of "recurrent" fibroadenomas in the left breast would suggest that this tumor alternatively represents a locally recurring benign phyllodes. However, as detailed earlier, there were no histologic findings to suggest this diagnosis. Additionally, scar tissue or changes suggestive of prior biopsy were not appreciated in a. Our patient had bilateral presentation of fibroadenomas, occurring synchronously at one point in time. Multiple fibroadenomas occur in about 15% of female patients, with equal proportions detected synchronously and metachronously in the same or opposite breast.¹ In contrast, multiple or bilateral fibroadenomas have not been

described in the male breast until now.

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Adjacent mammary parenchyma in the left excisional specimen of 2006. It has been speculated that hormonal imbalances, some due to medication use, cause proliferative changes in the male breast, such as gynecomastia, lobular differentiation, and fibroepithelial lesions. Although coexisting gynecomastia appears to be a consistent finding in male patients with fibroadenomas, the presence of lobular differentiation with or without associated gynecomastia is less common. Lobular differentiation was found in 2 reports of male to female transsexuals who were undergoing demasculinization and feminization via hormonal (ethinyloestradiol and ciproterone acetate) and surgical treatment.^{6,19} It is known that the slight increase of plasma estrogen to androgen ratio observed in idiopathic prepubertal or senile gynecomastia usually will not induce acinar and lobular formation in the male breast,^{4,5} but full acini and lobular formation will occur in transsexuals in whom progestagenic antiandrogens are combined with

feminizing estrogen therapy.^{6,7} Ciproterone acetate is such a progestagenic with a strong androgen receptor-blocking effect in the series reported by Ansah-Boateng and Tavassoli¹⁰; all patients also had concurrent gynecomastia. One patient was taking estrogen and another was taking methyl dopa chlorthalidone. Two patients were not taking any medications at the time of diagnosis. Nielsen¹¹ reported a case of bilateral fibroadenomatoid hyperplasia in a 69-year-old man who was taking digoxin, furosemide, and spironolactone. Both breasts had become diffusely enlarged with multiple fibroadenomatoid nodules during the last 4 years prior to presentation, when spironolactone was added; hence, it was surmised that this was the causative agent. Lastly, a 19-year-old woman with complete androgen insensitivity syndrome who was taking exogenous estrogen was reported to develop a juvenile fibroadenoma.¹² The presence/absence of lobular differentiation was not mentioned. Since gynecomastia is often coexistent in reported cases of fibroadenomas, it is difficult to discern whether exogenous drugs/medications leading to hormonal imbalances are causative of one or both lesions. To the best of our knowledge, there are no reports of fibroadenomas in male patients who did not have concurrent gynecomastia. The use of causative medications was not addressed in one case of a fibroadenoma in a 40-year-old man.²⁰ It is likely that in our patient, the long-standing use of

Lupron, a luteinizing hormone-releasing hormone agonist that is used to decrease testosterone levels for the treatment of advanced prostate cancer, contributed to the development of gynecomastia, lobular differentiation, and/or fibroadenomas. Breast enlargement is a known side effect of this medication.

References

- Rosen, P. P. Fibroepithelial neoplasms. In: Rosen PP, ed. *Rosen's Breast Pathology*. 2nd ed. Philadelphia, Pa: Lippincott Williams & Wilkins; 2001: 163–200.
- Holleb, A. I. , H. P. Freeman , and J. H. Farrow . Cancer of the male breast, part II. NY State J Med 1968. 68:656–663.
- Rosen, P. P. Benign proliferative lesions of the male breast. In: Rosen PP, ed. *Rosen's Breast Pathology*. 2nd ed. Philadelphia, Pa: Lippincott Williams & Wilkins; 2001:703–712.
- Nuttall, F. Q. Gynecomastia as a physical finding in normal men. J Clin Endocrinol Metab 1979. 48:338–340.
- Ribeiro, G. G. , H. V. Phillips , and L. G. Skinner . Serum oestradiol-17b, testosterone, lutenizing hormone, and follicle-stimulating hormone in males with breast cancer. Br J Cancer 1980. 41:474–477.
- Kanhai, R. C. , J. J. Hage , and E. Bloemena . et al. Mammary fibroadenoma in a male-to-female transsexual patient after hormonal treatment. Eur J Surg Suppl 2003. 588:69–71.
- Kanhai, R. C. , J. J. Hage , and P. J. van Diest . et al. Short-term and long-term histologic effects of castration and estrogen treatment on breast tissue of 14 male-to-female transsexuals in comparison with chemically castrated males. Am J Surg Pathol 2000. 24:74–80.
- Hilton, D. A. , J. S. Jameson , and P. N. Furness . A cellular fibroadenoma resembling a benign phyllodes tumour in a young male with gynaecomastia. Histopathology 1991. 18:476–477.
- Vancil, M. and W. Locke . Acromegaly, hyperparathyroidism, and probable mammary fibroadenoma in a man. Am J Surg 1965. 110:495–497.
- Ansah-Boateng, Y. and F. A. Tavassoli . Fibroadenoma and cystosarcoma phyllodes of the male breast. Mod Pathol 1992. 5:114–116.
- Nielsen, B. B. Fibroadenomatoid hyperplasia of the male breast. Am J Surg Pathol 1990. 14:774–777.
- Davis, S. E. and A. M. Wallace . A 19 year-old with complete androgen insensitivity syndrome and juvenile fibroadenoma of the breast. Breast J 2001. 7:430–433.
- Ng, W. K. , C. S. Poon , and My Lau . et al. Actin inclusions in stromal cells of fibroepithelial tumor of breast: immunohistochemical and ultrastructural studies. Ultrastruct Pathol 1999. 23:199–205.
- Pettinato, G. , J. C. Manivel , and E. W. Gould . et al. Inclusion body fibromatosis of the breast: two cases with immunohistochemical and ultrastructural findings. Am J Clin Pathol 1994. 101:714–718.
- Bittesini, L. , A. P. Dei Tos , and C. Doglioni . et al. Fibroepithelial tumor of the breast with digital fibroma-like inclusions in the stromal component: case report with immunocytochemical and ultrastructural analysis. Am J Surg Pathol 1994. 18:296–301.
- Hiraoka, N. , M. Mukai , and Y. Hosoda . et al. Phyllodes tumor of the breast containing the intracytoplasmic inclusion bodies identical with infantile digital fibromatosis. Am J Surg Pathol 1994. 18:506–511.
- Ortega, E. , F. I. Aranda , and M. T. Chulia . et al. Phyllodes tumor of the breast with actin inclusions in stromal cells: diagnosis by fine-needle aspiration cytology. Diagn Cytopathol 2001. 25:115–117.
- Otrakji, C. L. Fibroepithelial tumor. Am J Surg Pathol 1995. 19:730–732.
- Lemmo, G. , N. Garcea , and S. Corsello . et al. Breast fibroadenoma in a male-to-female transsexual patient after hormonal treatment. Eur J Surg Suppl 2003. 588:69–71.

Uchida, T. , M. Ishii , and Y. Motomiya
 . Fibroadenoma associated with gynaecomastia
 in an adult man: case report. Scan J Plast
 Reconstr Surg Hand Surg 1993. 27:327–329.

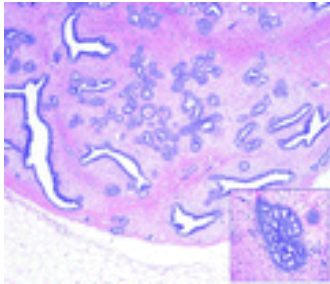


Figure-1-
 Fibroadenoma of the left breast excised in 2004. Low-power magnification exhibits the well-circumscribed contour of this fibroadenoma. Foci of florid duct hyperplasia (inset; hematoxylin-eosin, original magnification $\times 40$) and mildly cellular stroma are features reminiscent of juvenile fibroadenoma (hematoxylin-eosin, original magnification $\times 20$)

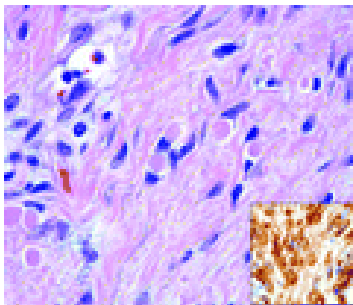


Figure-2-
 Stroma of fibroadenoma of the left breast excised in 2004. High-power magnification of the stromal component of this fibroadenoma demonstrates the presence of multiple, round, and brightly eosinophilic intracytoplasmic inclusion bodies identical to those seen in infantile digital fibromatosis (hematoxylin-eosin, original magnification $\times 400$). Present in stromal cells, these bodies are strongly positive for smooth muscle actin (inset; anti-smooth muscle actin, original magnification $\times 400$)

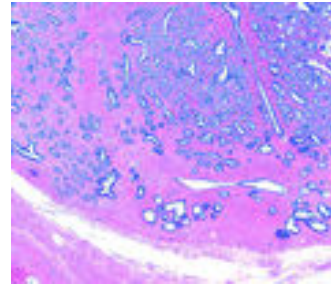


Figure-3-
 Fibroadenoma of the right breast excised in 2004. This low-power magnification exhibits the well-circumscribed tumor border of this fibroadenoma (hematoxylin-eosin, original magnification $\times 20$). Mildly atypical duct hyperplasia was focally identified (not shown)

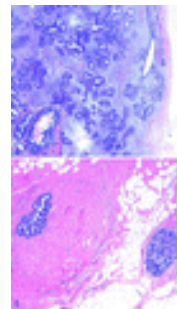


Figure-4-
 Fibroadenoma of the left breast excised in 2006. Low-power magnification demonstrates a well-circumscribed border, mildly cellular stroma, and a proliferative epithelial component (hematoxylin-eosin, original magnification $\times 20$). Prominent duct hyperplasia was focally atypical (inset; hematoxylin-eosin, original magnification $\times 200$). **Figure 5.** Right breast mass excised in 2006. Pathologically, the mass represented gynecomastia and focal lobular differentiation (hematoxylin-eosin, original magnification $\times 100$).

The authors have no relevant financial interest in the products or companies described in this article.

Conclusion –

A healthy male can grow fullblown breasts in pubertal age bilaterally[

gynaecomastia] which can show presence of fibroadenomas similar to female breasts.

Bilateral presentation of fibroadenoma with digital fibroma-like inclusions in the male breast.

Referances-

1. Sandra J. Shin, Paul Peter Rosen (2007) Bilateral Presentation of Fibroadenoma With Digital Fibroma-like Inclusions in the Male Breast. Archives of Pathology & Laboratory Medicine: Vol. 131, No. 7, pp. 1126-1129.
2. Arch Pathol Lab Med. 2007 Jul;131(7):1126-9.

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