

# Periductal Stromal Sarcoma of the Breast with Coexistent Tuberculous Mastitis

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### ABSTRACT

Periductal stromal sarcoma is a rare low-grade biphasic malignancy arising from periductal breast stroma. This tumor is distinct from phyllodes as it lacks the characteristic leaf-like architecture. Tuberculous mastitis is an uncommon infection seen rarely in the breast parenchyma. We present a rare association between the two diseases, which to the best of our knowledge is the first case reported so far.

**KEYWORDS:** Breast, periductal stromal sarcoma, tuberculosis

## INTRODUCTION

**P**eriductal stromal sarcoma (PSS) is a very rare neoplasm which arises from the connective tissue of the breast, especially the periductal stroma.<sup>[1]</sup> It has biphasic histology with a cellular sarcomatous stroma entrapping benign glands, however, lacking the phyllodes phenotype. Tuberculous mastitis is also an uncommon disease even in highly endemic countries. We here report a case of periductal stroma sarcoma with tuberculous mastitis in a 65-year-old lady. This is the first case report of such an unusual association of these two rare diseases.

## CASE REPORT

A 65-year-old lady presented to the surgical outpatient department (OPD) with lump in the right breast for the past 1 year. On examination, it was 4 cm × 2 cm swelling present in the right upper quadrant. It was hard and mobile. She had a history of excision of similar lump in a private hospital which was reported as fibroadenoma. The swelling recurred few months following excision. She gives no history of weight loss, cough, or contact with tuberculosis patients.

We received a lumpectomy specimen measuring 4 cm × 2 cm. Sections examined from it showed a hypercellular spindle cell lesion arranged in a vaguely fascicular pattern. The tumor cells showed oval to spindle nuclei with mild to moderate atypia and occasional conspicuous nuclei. Mitosis was 7/10 high-power field (hpf). Interspersed bland-looking breast ducts were identified surrounded by this cellular stroma

[Figures 1 and 2]. No broad papilla or leaf-like structure was seen. Necrosis was absent. The lesion was found to be reaching one of the resected margins. A possibility of PSS was given. The tumor cells showed focal positivity for CD34 on immunohistochemistry [Figure 3].

We received a mastectomy specimen of the same in due course. The mastectomy specimen measured 19 cm × 18 cm with attached axillary tail 5 cm × 4 cm. Nipple areola appeared unremarkable. A scarred area was seen in the upper outer quadrant indicative of the previous surgery. Sections examined from it showed multiple epithelioid cell granulomas with Langhans type giant cells surrounded by lymphocytes and plasma cells and with areas of caseation [Figure 4]. Stain for AFB was found to be positive. All of the thirteen axillary lymph nodes isolated also showed epithelioid cell granulomas. No caseation was, however, seen in the lymph nodes.

## DISCUSSION

PSS was considered a separate entity by the World Health Organization classification system at the consensus conference in Lyon in 2002 and is now separate from cystosarcoma with adipose metaplasia.<sup>[2-7]</sup> It occurs in pre- and post-menopausal women with a median of age of 55.3 years. The symptoms are similar to any benign or malignant breast disease.

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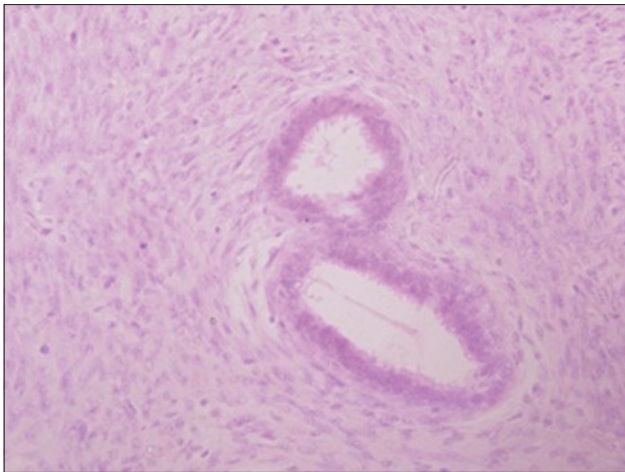
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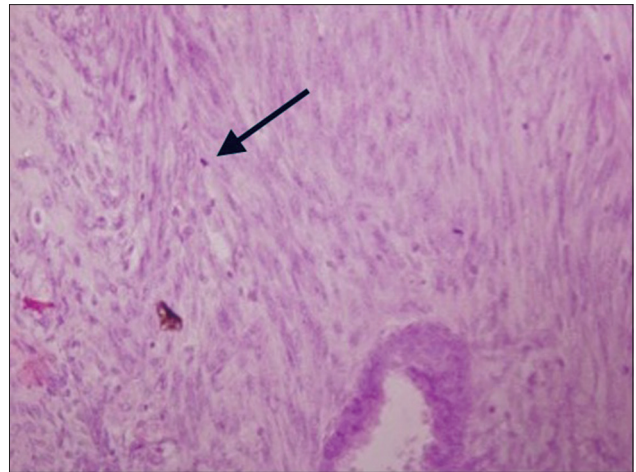


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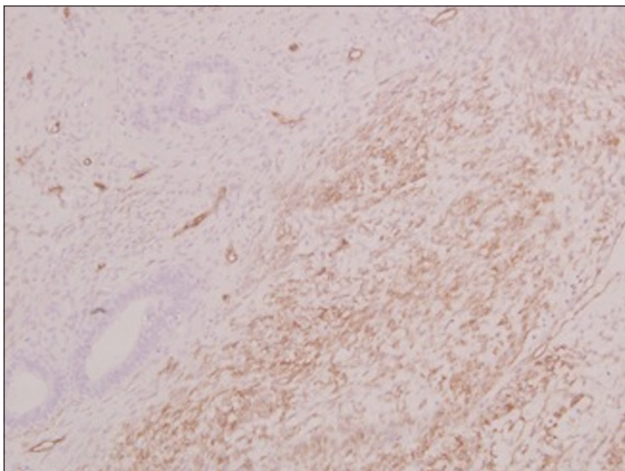
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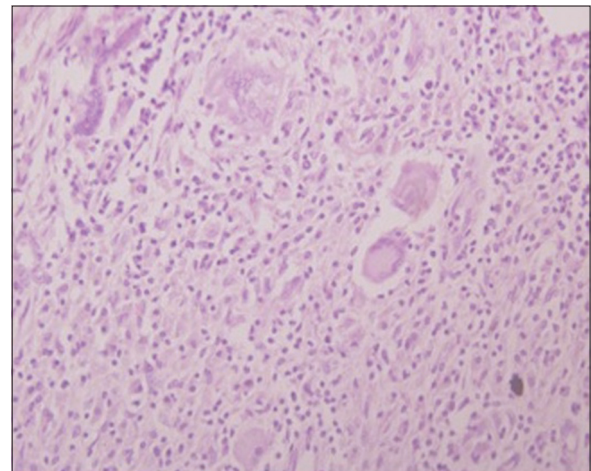
**Figure 1:** Photomicrograph of periductal stromal sarcoma showing bland breast duct surrounded by pleomorphic spindle cells (H and E, ×100)



**Figure 2:** Photomicrograph of periductal stromal sarcoma showing mitoses (black arrow) in the spindle cell stroma (H and E, ×400)



**Figure 3:** Photomicrograph showing focal CD34 positivity in stromal cells (IHC, ×100)



**Figure 4:** Photomicrograph showing epithelioid cell granulomas with Langhan's giant cells in breast stroma (H and E, ×400)

The histological features of PSS were defined by the Armed Forces Institute of Pathology<sup>[2]</sup> as under (a) a predominantly spindle cell stromal proliferation of variable cellularity and atypia around open tubules and ducts devoid of a phyllodes pattern; (b) one or, more often, multiple nodules separated by adipose tissue; (c) stromal mitotic activity of 3/10 hpf; and (d) infiltration into surrounding mammary fibroadipose tissue. Depending on the mitosis and the atypia, PSS can be graded from low to high grade. On immunohistochemistry, they express CD34 and lack S100, estrogen receptor, and progesterone receptor.

Our case demonstrated all of the above diagnostic criteria. A single nodule with prominent spindle cell proliferation was noted with mitotic activity more than 3/10 hpf. The lesion was reaching up to the resected margin. A focal positivity for CD 34 was also noted.

PSS is usually considered a tumor of intermediate grade and usually a wide excision is usually sufficient and

axillary lymphadenectomy is usually not required. Our patient underwent mastectomy due to her older age, recurrent nature of the lump, and the second lumpectomy being positive for the margins. Moreover, PSS has tendency to recur or eventually progress to phyllodes or soft tissue type sarcomas. It may occasionally exhibit intraepithelial changes ranging from ordinary hyperplasia to intraductal carcinoma. These warrant a close follow-up of such patients. The neoadjuvant chemotherapy or radiotherapy has not been found to be beneficial to such patients.

Mammary glands, spleen, and skeletal muscle are generally resistant to the tubercle bacilli survival and multiplication. Tubercular mastitis (TM) is thus rare among Indian population where tuberculosis is endemic. The incidence is known to range from 0.1% in developed countries to about 4% in India.<sup>[8,9]</sup>

TM is usually common in reproductive age group from 21 to 30 years and is rare in elderly patients. They can

even mimic carcinomas, especially in elderly patients. The occurrence of such rare diseases in a single patient has not been documented in literature. To our knowledge, this is the first case report of PSS with TM.

## CONCLUSION

The rare association of two very rare diseases is presented. Our patient was put on ATT and is on close follow-up in the OPD till the article was written and is doing fine.

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Nil.

## Conflicts of interest

There are no conflicts of interest.

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