Sclerosing lymphocytic lobulitis in the male breast

A H S Lee, B Zafrani, G Kafiri, S Rozan, R R Millis

Abstract
Sclerosing lymphocytic lobulitis is an inflammatory disorder of the breast that is well recognised in women. It has only been reported previously in two men; two further men with the condition are described here. Both presented with a breast mass, and one was an insulin dependent diabetic. Biopsy specimens from both patients showed circumscribed perivascular and, to a lesser extent, periductal collections of B and T lymphocytes. Sclerosing lymphocytic lobulitis in the female breast shows predominantly peribulbar inflammation. The predominantly perivascular distribution in men is consistent with the relative paucity of epithelium in the male breast. Interlobular fibrosis with epithelioid fibroblasts was also present. (J Clin Pathol 1996;49:609–611)

Keywords: breast, male, sclerosing lymphocytic lobulitis, inflammation.

Sclerosing lymphocytic lobulitis is a recently recognised disorder of the breast characterised by perilobular and perivascular aggregates of B and T lymphocytes, with increased expression of class II major histocompatibility antigens by the lobular and ductal epithelium, fibrosis and lobular atrophy. It is thought to be of autoimmune aetiology and is associated with other autoimmune diseases, particularly diabetes mellitus. The condition is well recognised in women, but has been reported in only two men. We describe two additional male patients in whom the perivascular and periductal inflammation has been characterised immunohistochemically.

Case reports
Patient 1 was a 47 year old Algerian man who presented with a 15 mm mass in the right breast. He had had insulin dependent diabetes mellitus for 31 years, but no history of other autoimmune diseases. Patient 2 was a 53 year old Greek man who presented with a poorly defined 5 cm breast mass. He did not have diabetes mellitus nor any other autoimmune disease, nor a family history of diabetes.

Pathological findings
An excision biopsy specimen was taken of the breast mass of both men. Sections from both
Figure 1  Plump fibroblasts were prominent in the stroma in patient 1.

Figure 2  A circumscribed lymphocytic infiltrate is present (A) around ducts and (B) around blood vessels.

showed male breast with no lobules. In patient 2 there was some epithelial hyperplasia and loose cellular periductal stroma consistent with gynaecomastia. In both biopsy specimens the interductal stroma showed fibrosis with plump fibroblasts with large oval nuclei that had an irregular outline and contained one or two small nucleoli. These fibroblasts were inconspicuous in patient 2 and prominent in patient 1 (fig 1). In both there was a circumscribed, well defined inflammatory infiltrate around blood vessels and, to a lesser extent, around ducts (fig 2).

The infiltrate was characterised by immunohistochemistry on paraffin wax sections using a Streptavidin biotin technique and the following primary antibodies: CD3 (T cells, 30 minutes microwave pretreatment; Dako, High Wycombe, UK); UCHL1 (CD45RO, primed T helper cells, macrophages, and a subset of B cells; Professor PCL Beverley, ICRF, London); L26 (CD20, B cells, 15 minutes microwave pretreatment; Dako); PGM1 (CD68, macrophages, 10 minutes trypsin; Dako); TAL-1B5 (α chain of HLA-DR, 15 minutes microwave pretreatment; ICRF, London).

The perivascular and periductal inflammation was composed of lymphocytes, with a slight predominance of T cells over B cells in patient 1, and predominance of B cells in patient 2. There were few macrophages. In each there was patchy staining of the ductal epithelium for HLA-DR.

Discussion

Sclerosing lymphocytic lobulitis is well recognised in women, but we have found only two previous reports of the condition in men. Both had longstanding insulin dependant diabetes mellitus and presented with a breast mass. Ashton et al emphasised the prominent lymphocytic vasculitis and less noticeable periductal lymphocytic infiltrate; Tomaszewski et al made no distinction between the changes seen in men and women. The inflammatory infiltrate was not characterised in the former report. In the latter the inflammation was composed predominantly of B cells, and there was negligible staining for HLA-DR with LN3. Both reports emphasised the presence of epithelioid fibroblasts in the fibrosis.

In both of our patients there was a perivascular and periductal lymphocytic infiltrate in a pattern quite similar to sclerosing lymphocytic lobulitis in the female breast, except that in women there is peribular rather than periductal inflammation. The predominantly perivascular distribution in men is consistent with the relative paucity of epithelium in the male breast. Although the inflammation was around the vessels, there was no vessel damage, thus it is not a vasculitis as suggested in previous reports. Plump fibroblasts were a prominent feature in patient 1, a diabetic man. It has been suggested that these fibroblasts are a marker of diabetic mastopathy, but they have also been reported in lymphocytic lobulitis not associated with diabetes. We have seen plump fibroblasts frequently in mammary stroma, and while they may be helpful in recognising sclerosing lymphocytic lobulitis, in our experience they are not a discriminatory feature. Although there was a slight predominance of T cells in one of our cases, B cells were prominent in both, a characteristic feature of sclerosing lymphocytic lobulitis. This is in contrast to most breast disorders where T cells, and to a lesser
Association between thyroid cancer of cribriform variant and familial adenomatous polyposis

K Hizawa, M Iida, T Yao, K Aoyagi, Y Oohata, R Mibu, K Yamasaki, T Hirata, M Fujishima

Abstract

A case of a 20 year old Japanese woman who developed thyroid cancer exhibiting unusual cribriform structures while being followed up for familial adenomatous polyposis/Gardner's syndrome is reported. The patient presented with osteomas, pigmented retinal lesions, and adenomas of the duodenum and the papilla of Vater, in addition to numerous adenomatous polyps in the colorectum. On ultrasonography, the thyroid cancer was localised to the right lobe and was identified as an irregular, internal echo tumour with a peripheral hypoechoic zone, measuring 1.8 cm in diameter. Histological examination of the resected tumour showed a concomitance of papillary proliferation and cribriform structures with follicles of varying sizes. These features can be distinguished from sporadic thyroid cancer.

Keywords: familial adenomatous polyposis, thyroid cancer.

Familial adenomatous polyposis (FAP)/Gardner's syndrome is an inherited disorder caused by a germline mutation of the APC gene. It is characterised by intestinal adenomatous polyps predisposing to cancer. Recent long term, follow up studies have clarified that patients with this condition are also susceptible to malignancies involving multiple sites. The thyroid gland, particularly in women, is reported to be the most frequently involved organ. Here, we present a young Japanese woman with thyroid cancer exhibiting unusual histological features of cribriform proliferation. These features are similar to those described recently by Harach et al. We also review previous cases described in the literature.

Case report

A 20 year old Japanese woman was admitted to hospital in July 1993 for treatment of colonic polyposis. The patient had been diagnosed as having familial adenomatous polyposis at the age of 11 years, when her mother had undergone a total colectomy for colon cancer involving adenomatosis of the colorectum. The patient's condition had been good and she had...