Case Report

An Uncommon Case of Diabetic Mastopathy in Type II Non-Insulin Dependent Diabetes Mellitus

Keiichi Sotome*1, Tatsuya Ohnishi*1, Ryo Miyoshi*1, Makoto Nakamaru*1, Akio Furukawa*1, Hiroshi Miyazaki*1, Kyoei Morozumi*1, Yoichi Tanaka*2, and Hisami Iri*3

*1Department of Surgery, Fussa Hospital, *2Division of Surgical Pathology Clinical Laboratory, Tokyo Dental College Ichikawa General Hospital, *3Department of Pathology, Fussa Hospital, Japan.

Diabetic mastopathy is an uncommon tumor-like proliferation of fibrous tissue of the breast that usually occurs in a patient who has suffered from type I diabetes mellitus of long duration. Here we report a rare case of diabetic mastopathy that occurred in type II non-insulin dependent diabetes mellitus. This patient was a 63-year-old postmenopausal woman. Mammography, ultrasonography and MR imaging could not distinguish it from breast cancer. Although the core needle biopsy specimen showed fibrosis without evidence of malignancy, excisional biopsy was performed. Histological findings demonstrated typical diabetic mastopathy with keloid-like fibrosis, perivascular lymphocytic infiltration, and lymphocytic lobulitis without evidence of malignancy. These lymphocytes were composed predominantly of B-cells. Five months after surgical biopsy, a nodular formation approximately 4 cm in diameter recurred adjacent to the resected end of the biopsy.


Key words: Diabetic mastopathy, Lymphocytic lobulitis, Diabetes mellitus, Type II non-insulin dependent

Diabetic mastopathy includes lymphocytic ductitis and lobulitis with varying degrees of keloidal fibrosis, vasculitis, epithelioid fibroblasts, and lymphoid nodule formation1-4). Until recently, most researchers have reported that diabetic mastopathy usually occurs in patients who have suffered from type I diabetes mellitus of long duration4-7). Although there have been some reports of diabetic mastopathy in a patient with type II diabetes mellitus, there have been few cases in non-insulin dependent patients3, 8-12). Here we report an unusual case of diabetic mastopathy in a type II non-insulin dependent patient. In this report, we also review MR findings that have been described in the literature10, 11, 13, 14).

Case Report

A 63-year-old postmenopausal woman was affected by type II diabetes mellitus. An oral diabetic agent had been given since the age of 53, resulting in a HbA1c in the range of 6.2-7.5% without insulin therapy. She had previously suffered from diabetic neuropathy. She noticed a mass in her left breast about three years previously which had gradually enlarged. The left breast was about 4 cm in diameter when she was referred to our hospital at the age of 63. The mass was hard, ill-defined and very movable without any skin symptoms such as dimpling, thickening or peau d’orange. There was no axillary lymphadenopathy. Mass, microcalcification and architectural distortion were not seen on mammography (Fig 1). Ultrasonography of the lesion revealed a heterogenous hypoechoic mass with an indistinct margin, suggesting breast cancer (Fig 2). MR imaging of the lesion revealed a blurred contrast enhanced area, and from that finding malignancy could not be excluded (Fig 3). The specimens obtained from the mass by ultrasound-guided core needle biopsy showed fibrosis without any malignancy. Although there was no definite change in size, hardness or shape of the mass after three months of follow-up, she requested resection of the mass. Almost the whole lesion was removed by excisional biopsy. Histological findings demonstrated keloid-like fibrosis, lymphocytic lobulitis, and...
perivascular lymphocytic infiltration without evidence of malignancy (Fig 4A, B, C). These lymphocytes were composed predominantly of B-cells since they were positive for CD20 (clone L26, Dako Cytomation, Glostrup, Denmark), which identifies mature B-cells, and negative for CD3 (clone PS1, Novocastra, Newcastle, UK), which identifies T-cells (Fig 5A, B). Five months after surgery, a nodular formation approximately 4 cm in diameter similar to the previous lesion appeared adjacent to the resected end of the biopsy. Ultrasonography and MR imaging of the lesion showed findings similar to those of the initial examination. Core needle biopsy of the lesion was performed again and fibrosis was detected without any malignancy. In April 2005, there were no further changes in the clinical characteristics, except for a slight increase in the size of the lesion.

Discussion

Diabetic mastopathy is an uncommon tumor-like proliferation of fibrous tissue of the breast, mimicking breast cancer clinically, and usually occurs in patients with type I diabetes mellitus of long duration. Since Soler and Khardori first reported 2 cases of fibrous disease of the breast in association with longstanding type 1 diabetes mellitus in 1984, 168 cases of diabetic mastopathy that occurred in patients with type I diabetes mellitus have been reported in 29 papers so far. Although it is rare, the condition has also been observed in patients with type II diabetes mellitus. Twenty-five cases of diabetic mastopathy that occurred in type II diabetes mellitus have been reported in 10 papers. Among them, 10 out of 25 patients had type II non-insulin dependent diabetes. Thus, our case of diabetic mastopathy in a patient with type II non-insulin dependent diabetes is rare.

Pathological findings of diabetic mastopathy were described in detail by Tomaszewski et al. and Seidman et al. These include keloid-like fibrosis, lymphocytic perivasculitis, mononuclear...
ductitis and lobulitis. Although epithelioid fibroblasts were described as an integral component of diabetic mastopathy by Tomaszewski et al., Seidman et al. reported that epithelioid fibroblasts were unnecessary for the diagnosis of diabetic mastopathy. Our case had diabetic mastopathy by histological examination of a surgical biopsy specimen showing keloid-like fibrosis, perivascular lymphocytic infiltration, and lymphocytic lobulitis. Epithelioid fibroblasts were not detected. Epithelioid fibroblasts were detected in 5 out of 10 reported cases of diabetic mastopathy in type II non-insulin dependent diabetes so far (8, 10). Seidman et al. also stated the lymphocytic infiltration in diabetic mastopathy consists primarily of B cells, in contrast to non-diabetic mastitis in which lymphocytic infiltration consists primarily of T cells (8). Our case also showed B cells.

Generally, diabetic mastopathy is a hard, irregular, movable, nontender, single or multiple, unilateral or bilateral mass (8, 10). Mammography shows the presence of a dense parenchymal structure with no distortions or microcalcifications (8). Ultra-

**Fig 4.** A: Histological findings revealed dense keloid-like fibrosis, lymphocytic ductitis, and lymphocytic lobulitis. B: Lymphocytic lobulitis, background in keloid-like fibrosis. C: Perivascular lymphocytic infiltration, background keloid-like fibrosis.

**Fig 5.** A: Lymphocytic infiltrate showing predominance of CD20 positive mature B-cells. B: CD3 positive T-cells were not detected in the cellular infiltrates.
sound findings vary from irregular hypoechoic mass with marked acoustic shadowing to a vague hypoechoic area without shadowing. These appearances are nonspecific and mimic those of scirrhous or invasive lobular carcinoma. MR imaging findings, although there are only a few reports, vary from no enhancement to slight heterogeneous enhancement of the mass. In our case, MR imaging revealed a blurred contrast enhanced area, which is not typical of malignancy, but malignancy could not be excluded.

Ultimately, in order to diagnose diabetic mastopathy, histologic confirmation is necessary. To determine whether this lesion could be diagnosed as diabetic mastopathy without surgical biopsy, we re-examined three core needle biopsy specimens retrospectively. In these specimens, there were findings of fibrosis, but not keloid, a focus of perivascular lymphocytic infiltration, and some foci of lymphocytic lobulitis without evidence of malignancy. If we had been aware of the entity of diabetic mastopathy, we might have been able to make the correct diagnosis.

In our case, a nodular formation appeared adjacent to the resected end of the biopsy 5 months after surgery. Recurrence of diabetic mastopathy was proved by core needle biopsy specimens from the nodule which had the same histological findings as the specimens of the initial biopsy. With respect to the recurrence of diabetic mastopathy after surgical examination, Camuto et al. reported that since recurrences tended to be in the same location and involved more breast tissue than the preceding lesion, it is possible that the mastopathy affects the healing process and that surgery itself exacerbates the condition.

In conclusion, we should be aware of diabetic mastopathy when we encounter breast mass in patients with diabetes mellitus. Because the findings of diabetic mastopathy by physical examination, ultrasonography and MR imaging are nonspecific and mimic those of breast cancer, histological examination by core needle biopsy is required. If typical features of diabetic mastopathy exist, surgical biopsy should be avoided because of the tendency for recurrence.

References


