Membranous-Lipodystrophy-Like Changes among Hemodialysis Patients with Carpal Tunnel Syndrome

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Dear Sir,

Membranous lipodystrophy is the name previously given by Nasu et al. [1] to a rare disease in which cyst-lesions of adipose tissue, including that of long bones, occur together with sudanophilic leukoencephalopathy. This disease has recently been recognized as hereditary and is characterized by polycystic changes in many bones, with a variety of central nervous system changes due to impairment of lipid metabolism by an unknown mechanism [2-6]. Histopathological diagnosis is possible by biopsying bone marrow or synovial tissue. The adipose cells of the bone marrow, synovial membrane and other sites are replaced by convoluted, hyaline, eosinophilic membranes of variable thickness that enclose large spaces. The membranes are positive for periodic acid-Schiff staining but negative for resorcinol-fuchsin staining for elastic tissue. Masson's trichrome and Azan-Mallory stain the membranes blue or red. In frozen sections, the membranes and the contents of the cyst-like spaces are Sudan III positive [7].

We have found exactly the same membranocystic changes in subcutaneous tissue close to arteriovenous shunts in 93 of 100 cases in hemodialysis patients with carpal tunnel syndrome. The average duration of hemodialysis was 12.6 years, and the average age of the patients was 52.2 years (male:female = 54:39). These changes were quite

abundant in the synovial tendon sheaths (89.5%) but minimal in the tendons (< 1%).

Conventional histochemical studies revealed an eosinophilic hyaline substance in all synovial tendon sheaths. Most of the membranocystic lesions contained material positive for Masson's trichrome stain (fig. 1a). The membranes of the membranocystic changes appeared markedly convoluted, and the thickness of the membrane varied from lesion to lesion. As summarized in table 1, they stained red with Masson's trichrome, were periodic acid-Schiff positive, stained blue with Luxol fast blue, were Sudan black B positive but negative for Alcian blue (pH 1.0) and the resorcinol-fuchsin stain. The tissues did not stain with Congo red. No β 2-microglobulin was found by the peroxidase-antiperoxidase method. Interestingly, they reacted with antisera against amyloid P component and neuraminidase. Occasional macrophage-like cells were present

in the tissues. It was confirmed by electron microscopy that the membranocystic lesions were the same as those found in membranous lipodystrophy. The lesions showed membrane undulations that contained a substance similar to the fat droplets of adipose cells (fig. 1b).

It has been shown that membranous-lipodystrophy-like changes can be produced by several forms of circulatory disturbances and that they are among the nonspecific

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Fig. 1. a Membranocystic lesion found in adipose tissue around arteriovenous shunt of hemodialysis patient with carpal tunnel syndrome. Masson's trichrome. Bar = 50 μm . b Electron micrograph of membranocystic lesion. Note the variously shaped membrane structures and the marked development of microtubules. Bar = 5 μm .

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Table 1. Histochemical properties in membranocystic lesions of hemodialysis patients with carpal tunnel syndrome (A) compared with those in secondary amyloidosis due to $\beta 2$ -microglobulin (B) and membranous lipodystrophy (C)

changes of adipose tissue. Membranocystic lesions in adipose tissue similar to those of membranous lipodystrophy have been reported in several conditions other than membranous lipodystrophy, such as lupus erythematosus profundus [8] and limb necrosis caused by chronic arterial obstruction [9]. It is of interest that circulatory disturbance due to arteriolar and capillary necrosis were originally considered responsible for the membranocystic lesions of membranous lipodystrophy. Thus, the membranocystic lesions of hemodialysis patients with carpal tunnel syndrome may represent nonspecific changes of adipose tissue resulting from circulatory impairment around arteriovenous shunts.

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