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Mushroom-like soft fibromas on chronic leg lymphedema

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Mushroom-like soft fibromas on the chronic lymphedema of the legs.

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The patient gives written informed consent for publication.

Dear Sir,

We present an unusual case of soft fibromas on the legs with lymphedema seen in a diabetic woman. Only two preceding cases have been reported.¹⁻³⁾ Comparison of the cases disclosed many common features of this condition.

A 47-years-old woman noticed a hot sensation on her bilateral legs in July 2004. She had leg edema of two years duration and yellowish normal skin colored tumor for 1 year. She had systemic lupus erythematosus for 22 years and corticosteroid-induced diabetes mellitus for 10 years. She had also lupus nephritis and was treated by prednisolone (20-30mg) and cyclophosphamide for 9 years. No surgical procedure, which may be related to leg edema, was documented. Physical examination revealed that her weight was 125kg; height was 160cm; and her temperature was 38.9C. Her legs were markedly swollen with diffuse erythema. There were up to 2-3cm sized, partially violaceous, yellow-skin colored elastic soft nodules and tumors on bilateral legs with no exudation. Inguinal lymph nodes were not palpable. Complete blood count showed white blood cells, 11,400/mm³ (normal; 3000-7800/mm³); hemoglobin, 10.4g/dl (normal; 10.4-14.4g/dl); total protein, 4.8g/dl (normal; 6.5-8.0g/dl); and albumin, 3.0g/dl (normal; 4.0-5.2g/dl). Liver function tests were mostly normal. BUN and creatinine were raised to 51.2mg/dl (7.0-24.0mg/dl) and 3.65mg/dl (0.4-0.9mg/dl), respectively. C-reactive protein showed 29.94mg/dl (<0.5mg/dl). She admitted to our hospital under the diagnosis of cellulitis with lymphedema, and antibiotics with γ -globulin therapy were initiated for the purpose of control for severe infection with good response.

She visited with similar episode of cellulitis in April, May and July 2005 (Fig. 1a). During the course, yellowish normal skin-colored mushroom-like tumors were increased and enlarged on her legs. The tumors were more prominent on her right leg and were not seen on thigh.

Because her diabetes-induced renal failure exacerbated, hemodialysis therapy was initiated in Jan. 2006. This resulted in the improvement of her swollen legs accompanied by the decrease in soft fibromas. Her prednisolone dose was decreased to 12.5mg/day and in Sep. 2007, the fibromas were considerably decreased (Fig. 1b). Prednisolone was more decreased to 5mg /day in Mar 2008. She had no cellulitis during the last three years and the fibromas were mostly gone.

The histopathology of the tumor disclosed fibroblasts with no atypia, vascular dilation and proliferation, and severe stromal edema with mild inflammatory infiltrates (Fig. 1c). Beneath the atrophic epidermis, vascular dilation with peripheral lymphocytic infiltrates and extravasations were seen. We diagnosed the tumor as a kind of soft fibroma.

Recent report indicates that angiogenesis is induced in inflammation with or without vascular endothelial growth factor (VEGF).⁴ In our case, recurrent cellulitis associated with leg edema might have induced this type of peculiar soft fibromas.

This ‘mushroom-like’ peculiar soft fibroma¹ was first described in 1983 by Huntley², and after that, only one case report has been published.³ Our case is the 3rd one with long term observation. Common manifestations of the cases are 1) diabetes- or obesity-related edema of the legs, 2) hypervascular condition (recurrent cellulitis or Morbus Kaposi), and 3) hyper-corticosteroid condition (Cushing’s syndrome and prednisolone therapy).

Interestingly, the tumors were improved following hemodialysis and tapering of prednisolone dose. Recurrent cellulitis subsided and soft fibromas were decreased in number and size (compare Fig. 1 a and b). Long-term observation disclosed the spontaneous resolution indicating the dynamic nature of this peculiar soft fibroma.

Because of the unique clinical presentation, we named this as “mushroom-like soft fibromas on the chronic lymphedema of the legs”

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Legends to Figures

Fig 1a

Clinical presentation of the skin tumors.

The lesions were up to 2-3cm sized, partially violaceous, yellow-skin colored, elastic soft tumors and nodules. These tumors are enlarged and increased in number, and represented as mushroom-like (Jul. 2005).

Fig 1b

Almost all fibromas are subsiding. Most but not all the tumors are gone. (Sep. 2007).

Fig 1c

Histopathological findings (Hematoxylin-Eosin stain).

Low magnification (X40).

Dome-shaped pedunculated tumor.

Inflammatory infiltrate is observed. Hyper-vascularity and dilated vessels are noted.





