A Case of Eosinophilic Panniculitis Associated With Superior Sagittal Sinus Thrombosis

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Eosinophilic panniculitis is characterized by a prominent infiltration of numerous eosinophils in subcutaneous fat, and has been identified in patients with a variety of associated clinical conditions. A case of eosinophilic panniculitis in a 20-year-old woman with a history of atopic dermatitis is reported. She later developed superior sagittal sinus thrombosis, and we stress the importance of systemic evaluations in patients with eosinophilic panniculitis.
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Key Words : Eosinophilic panniculitis, Superior sagittal sinus thrombosis .

Eosinophilic panniculitis is characterized by a prominent infiltration of numerous eosinophlils in subcutaneous fat, and may be considered as a reactive condition because most patients have associated systemic conditions including arthropod bites, infections, drug injections, and other immunoreactive disorders[1].

We report a case of eosinophilic panniculitis with characteristic clinicopathologic findings associated with superior sagittal sinus thrombosis which had not been reported until now.

CASE REPORT

A 20-year-old, previously healthy woman presented with two palm-sized hard and tender plaques on the right lower back and shin. They had been present for 10 days and the patient complained about them feeling hot. The eruption first occurred on her back, preceded by an erythematous induration which she had noticed at the site of an insect bite, but within a few days, the lesion had gradually spread forming a tender plaque and a similar lesion developed on the shin. She therefore visited the department of internal medicine and was referred to our department.

A clinical examination revealed an erythematous, tender, indurated area with a peau d'orange appearance on the right side of the lower back and shin, measuring approximately 10×8 cm and 5×4 cm, respectively. A central puntum or evidence of an arthropod bite was not found within the lesions. There were no constitutional symptoms or preceding illnesses except for mild atopic dermatitis which she had had since infancy.

A skin biopsy from the back showed sparse infiltration of eosinophils, lymphoid cells and histiocytes around superficial and deep dermal vessels and eccrine sweat glands. Strikingly dense infiltration almost exclusively composed of eosinophils that affect both septa and lobules was noted on the panniculus. Eosinophils were scattered between collagen bundles of the reticular dermis, but typical flame figures of eosinophilic cellulitis were not seen. There was no evidence of vasculitis.

Laboratory studies revealed leukocytosis of 15,500/mm^3 with 38.0% eosinophils, absolute eosinophilia of 5,200/mm^3, and thrombocytopenia of 95,000/mm^3. Other values including routine

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Table. Associated conditions in patients with eosinophilic panniculitis*

Arthropod assault
Dermatitis
Atopic dermatitis, Contact dermatitis
Drug injection
Erythema nodosum
Hypereosinophilic syndrome
Infection
Gnathostoma, Toxocara, Streptococcus
Injection granuloma
Leukocytoclastic vasculitis
Lupus panniculitis
Malignancy
Cervical carcinoma, Lymphoma, Rectal carcinoid, Refractory anemia with excess of blasts, Eosinophilic leukemia
Morphea profundus
Wells' eosinophilic cellullitis

* Modified from J Am Acad Dermatol. Vol 34 : 229-234, 1996.

hematological and chemistry profiles and autoimmune studies were all normal or negative. Antibiotics (levofloxacin) and non-steroidal antiinflammatory drugs (talniflumate) were administered for 3 days but improvement was not seen and she complained of nausea and headaches. So the antibiotics were changed to cefaclor and dexamethasone injections were given intramuscularly. After 4 days, the skin lesions showed a slight improvement with loss of pain and tenderness but headaches and vomiting persisted. On the tenth day after her initial visit, she developed severe headaches, vomiting, motor weakness on the right side and aphasia. A brain CT showed a hemorrhagic transformation of an infarct in the left temporoparietal lobe. She had an emergency operation for hematoma removal and a further work-up to find out the nature of the cerebral lesion was performed. No tumorous lesion nor parasites were found in the imaging studies of the brain. A bone marrow biopsy and peripheral blood smear were all normal. VDRL, FANA, Scl-70, Anti-RNP, Anti-Sm, antiplatelet antibodies and anticardiolipin antibodies were all negative. MRI and 4-vessel-angiography of the brain revealed thrombotic obstruction of the superior sagittal sinus and infarction of the left frontoparietal lobe. Finally she was diagnosed as having superior sagittal sinus thrombosis and was treated with surgery and antithrombotic, anticonvulsive medications. The initial skin lesions resolved without scarring or atrophy within a few weeks. However, after 6 months, one episode of recurrence was noted in the opposite leg without accompanying intracranial thrombosis. The recurrent lesion disappeared after about ten days without specific treatment and during a 3-month follow-up period she was free of the skin lesions.

DISCUSSION

The term 'eosinophilic panniculitis' was initially coined by Burket and Burket in 1985[2], in a patient with distinctive panniculitis that had the histological changes of Well's syndrome (flame figures in the dermal-subcutaneous junction). They considered these pathological alterations so unique and proposed to extend the concept of Well's eosinophilic cellulitis to the lesions of the panniculus. Since then, other cases showing similar histopathological changes in the panniculus have been described and it has been considered not a specific disease entity but a unique reactive process of the patients associated with various inflammatory and immunoreactive diseases[1,3-7].

The clinical spectrum of lesions that display pathological changes of eosinophilic panniculitis is diverse. Nodular lesions are the most common but plaques, papulovesicles, macular lesions, purpura, erosive lesions, and dermal processes including angioedema, urticaria also occur[3]. The most char-

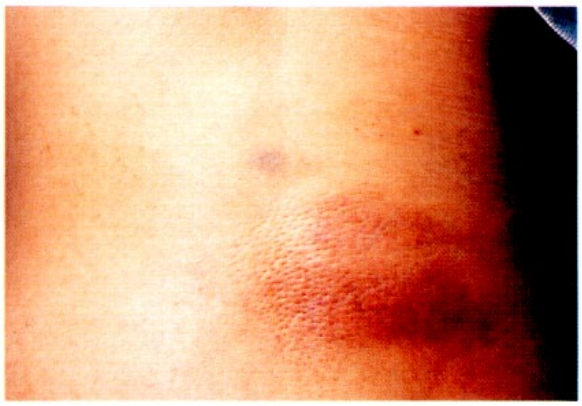

Fig. 1. Erythematous plaque with peau d'orange appearance on the right lower back.

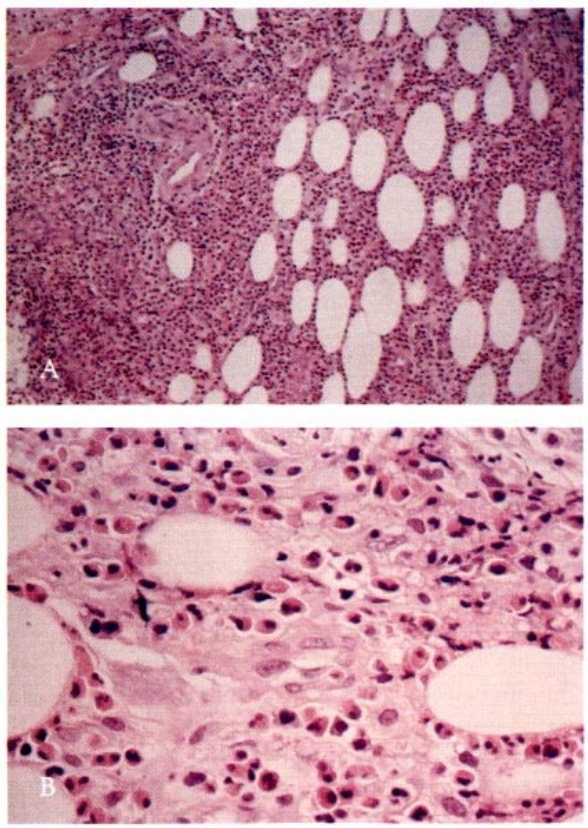

Fig. 2. A. Dense infiltration of eosinophils in the reticular dermis and panniculus(×100). B. High power view shows infiltration almost exclusively consisting of eosinophils in the panniculus (×400).

acteristic histopathological finding is infiltration of numerous eosinophils involving both septa and lobules of the subcutis[1-4] with fewer lymphocytes and macrophages. Some patients had pathological changes of eosinophilic cellulitis, characterized by flame figures, within their leisons[2,3,5] but others did not [1,6,7].

Eosinophilic panniculitis may be considered as a reactive process because most patients have an associated systemic condition(Table)[1,3,4]. In the reported cases, the most commonly associated diseases are gnathostomiasis, leukocytoclastic vasculitis, erythema nodosum and arthropod bites. Therefore, systemic evaluation of a patient with eosinophilic panniculitis should be performed based on the potentially associated conditions. They include a detailed history, complete physical examination and routine laboratory studies and subsequent evaluations should be based on the result of preliminary findings. The underlying associated conditions of eosinophilic panniculitis may not be apparent at the time of initial evaluation, therefore continued surveillance is necessary[1].

Eosinophilic cellulitis has a close relationship to eosinophilic panniculitis[3]. There may be infiltration of eosinophils also in the panniculus of Wells' syndrome, but in most cases histological changes were limited in the dermis with typical flame figures and rarely involving the subcutaneous fat[1,2]. In our patient, clinically tender erythematous plaques suggestive of eosinophilic cellulitis or erythema nodosum were present. Diagnosis of eosinophilic cellulitis should be considered because even in eosinophilic cellulitis, flame figures can only be detected in acute and subacute stages and may be quite rare in number even in these stages. However, in our case, histopathological changes mostly limited to the panniculus and the absence of flame figures or other dermal changes of typical eosinophilic cellulitis led us to the diagnosis of eosinophilic panniculitis.

Other eosinophilic infiltrative processes of subcutis in the differential diagnosis of eosinophilic panniculitis include Chrug-Strauss allergic granulomatosis, eosinophilic fasciitis, hypereosinophilic syndrome and erythema nodosum[5]. Chrug-Strauss allergic granulomatosis is a granulomatous vasculitis and pulmonary involvement is prominent. Eosinophilic fasciitis clinically resembles eosinophilic panniculitis, but it shows scleroderma-like induration and fascial involvement may cause muscle groups to be separated by a line of demarcation (groove

sign), and the veins may appear depressed (sunken veins)[8]. Histologically, eosinophilic fasciitis is characterized by fibrosis of the superficial fascia and fibrous septa of the subcutis. Eventually the adipocyte lobules are replaced by thick collagen bundles. The infiltrate of eosinophils is variable in intensity. The hypereosinophilic syndrome has been reported to involve the skin in approximately 50% of patients[9]. The lesions are not pathognomonic, but generally consist of pruritic macules and papules, or urticaria and angioedema accompanied by a persistent marked periphral blood eosinophilia. The histology is that of a perivascular dermal eosinophilic infiltrate. Erythema nodosum is clinically similar to eosinophilic panniculitis; however, eosinophils are not prominent and the panniculitis is primarily septal.

Our patient later developed superior sagittal sinus thrombosis resulting in intracerebral hemorrhage and infarction. Antiphospholipid antibody syndrome or other immunologically mediated syndromes causing venous thrombosis were suspected, but systemic evaluations revealed no abnormalities. There had been no report of eosinophilic panniculitis associated with venus thrombosis or other disorders of blood coagulation and we considered this as a first case. Although thrombosis of intracranial blood vessels developed after the initial lesions of eosinophilic panniculitis, the underlying associated conditions may not be apparent at the time of initial evaluation1. Therefore continued surveillance is warranted.

Reports of eosinophilic panniculitis are rare and only one case is presented in the Korean dermatologic literature[10]. We report a rare case of eosinophilic panniculitis associated with superior sagittal sinus thrombosis for the first time and stress the importance of systemic evaluation in patients with eosinophilic panniculitis.

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