Post herpes-zoster scar sarcoidosis with pulmonary involvement

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ABSTRACT

Cutaneous sarcoidosis presents with a wide range of clinical presentations. An uncommon cutaneous manifestation is infiltration of old cutaneous scars with non-caseating granulomas known as scar sarcoidosis. Most of the patients with this clinical entity have other systemic manifestations, particularly pulmonary changes. We report a case of a 50 years old man, presenting with cutaneous sarcoidosis overlying scars of healed herpes zoster.

Key words: Herpes zoster, pulmonary sarcoidosis, scar sarcoidosis

INTRODUCTION

Scar sarcoidosis, a rare presentation of cutaneous sarcoidosis, refers to infiltration of old scars with non-caseating epithelioid cell granulomas.[1] Scar infiltrates may appear early in the disease, before pulmonary parenchymal changes and may be associated with hilar adenopathy or erythema nodosum. Most patients with scar sarcoidosis have other systemic manifestations and changing scars in patients in remission may indicate disease exacerbation.[1]

CASE REPORT

The present case report is about a 50-year-old man who presented with multiple, mildly tender skin colored lesions over left upper back of 18 months duration. Patient had a history of eruption of painful, grouped blisters in this region 2 years before, that resolved leaving behind scars. At 6 months later, patient noticed skin colored raised lesions appearing over these scars that gradually increased in extent. He also complained of low grade fever and progressive breathlessness on routine work for last 6 months. There was no history of hemoptysis. Cutaneous examination revealed multiple irregular, grouped skin colored papules and plaques of 3-10 mm in size arranged in a zosteriform pattern along right thoracic (T1) segment [Figure 1]. There was no evidence of excessive scarring or keloid formation at the site of previous healed injuries elsewhere in the body. Systemic examination including chest examination did not reveal any significant finding. Hematological investigations including serum calcium were within the normal limits. The level of serum angiotensin-converting enzyme was 82 μg/L (normal range 8-47 μg/L). Ophthalmologic examination and electrocardiogram were normal. Mantoux test performed with 1 IU of PPD was negative after 48 h. Sputum for acid fast bacilli (AFB) was negative on 3 consecutive occasions. Chest radiography showed hilar prominence on the right side. Contrast enhanced computed tomography – chest revealed bilateral lung fields showing miliary nodules along with subpleural involvement with interlobular peribroncho-vascular thickening and interspersed areas of consolidation involving bilateral upper lobes, lingula and apical segment of right upper lobe. Areas of fibrosis in right upper lobe with emphysematous changes were seen along with bilateral hilar lymph nodes [Figure 2]. The findings

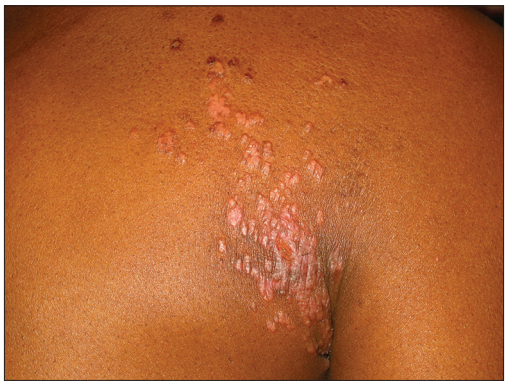

Figure 1: Multiple irregular, grouped skin colored papules and plaques arranged in a zosteriform pattern along the right thoracic (T1) segment

Access this article online

Website: www.idoj.in

DOI: 10.4103/2229-5178.126041

Quick Response Code:

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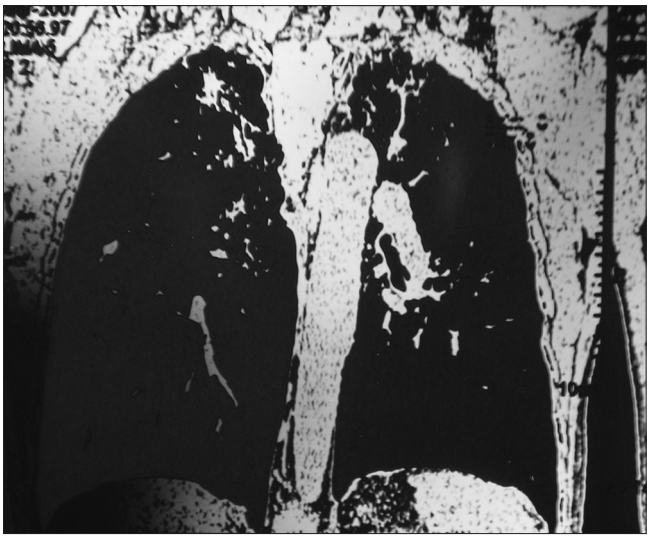

Figure 2: Contrast enhanced computed tomography chest showing miliary nodules along with subpleural involvement with interlobular peribroncho-vascular thickening and interspersed areas of consolidation

were suggestive of pulmonary sarcoidosis. Ultrasound abdomen was normal.

Histopathological examination of lesional biopsy on hematoxylin-eosin stained sections revealed multiple, non-caseating epithelioid granulomas with Langhan's giant cells, focal fibrinoid necrosis and occasional lymphocytes at the periphery in superficial and deep dermis consistent with the diagnosis of scar sarcoidosis [Figure 3]. The evidence of scar was evident in the form of haphazard thick collagen bundles in the dermis extending up to the subcutaneous tissue. Polarizing microscopy and special stains for AFB and fungus were negative. A diagnosis of scar sarcoidosis with pulmonary involvement was made. Patient was prescribed prednisolone 40 mg/day and clobetasol propionate (0.05%) cream to be applied twice a day over the lesions. At 4 weeks treatment led to decreased breathlessness and a 50% reduction in size of the lesions.

DISCUSSION

Sarcoidosis is a chronic multi-systemic granulomatous disease of unknown etiology. All body parts can be involved, but the organ most frequently affected is lung. Involvement of skin, eye, liver and lymph nodes is also common. Cutaneous involvement in sarcoidosis occurs in 10-35% of patients with systemic disease.[1]

Scar sarcoidosis is a rare but specific form of cutaneous sarcoidosis in which old scars become infiltrated with non-caseating epitheloid cell granulomas. The exact incidence of scar sarcoidosis is not known and is reported to vary from 2.9% to 13.8% in series of adult cutaneous sarcoidosis.[2]

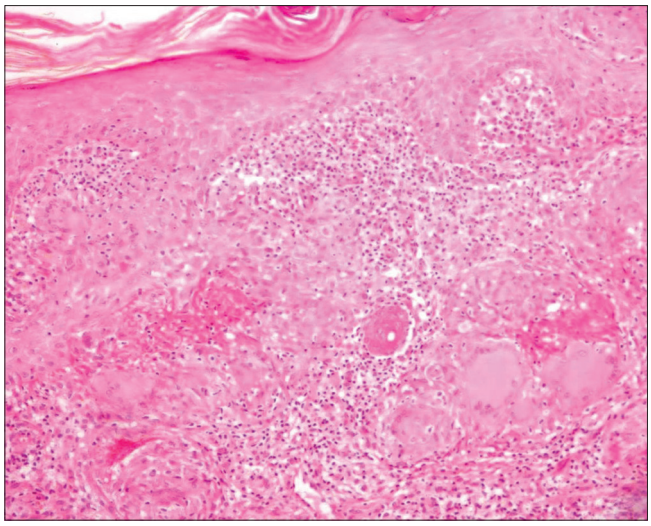

Figure 3: Photomicrograph showing granulomas with epitheleiod histiocytes, Langhan's giant cells and central fibrinoid necrosis (H and E, ×100)

In addition to reactivation of scars obtained from previous wounds, it has been reported at the sites of previous intramuscular injections, blood donation puncture sites, tattoo scars, scars of herpes zoster, on ritual scarification, at the sites of allergen extracts for desensitization and following hyaluronic acid injection and laser surgery.[3,4] Scar sarcoidosis may be isolated or precede or accompany systemic sarcoidosis, most often pulmonary sarcoidosis. Changes in scars in patients with sarcoidosis in remission may indicate exacerbation of disease or may even be a marker of recurrence of thoracic sarcoidosis.[5]

Scar sarcoidosis appearing on post-herpes zoster scars have been reported on few occasions [Table 1].[4-8] It has been postulated that this hypersensitivity induced granulomatous tissue reaction encountered in the healing phase of herpes zoster lesions depends on the patient's immune status and presence of viral antigens or tissue antigens modified by the varicella-zoster virus. Of the 5 previously reported cases only two had systemic involvement in the form of pulmonary disease. There is a debate regarding nomenclature of the entity scar sarcoidosis without systemic involvement. Few authors have suggested that on scars of herpes zoster, this entity should be termed sarcoidal granuloma akin to isotopic response.[7]

As many of the patients have spontaneous remissions and the benefits of therapy do not affect the long-term outcome, systemic treatment is usually not indicated for cutaneous lesions, unless they are progressive and disfiguring and is reserved for patients with severe or progressive pulmonary symptoms. Intralesional corticosteroids and high potency topical corticosteroids are beneficial for cutaneous lesions. Excision of small lesions has been reported to be successful, although there is the risk of inducing sarcoidal infiltrates. Alternative therapies include anti-inflammatory and immunosuppressive

Table 1: Details of patients with post herpes zoster scar sarcoidosis

Author/year	Age/ sex	Site	Duration of lesions	Systemic involvement
Bisaccia *et al* (1983)[4]	52/F	Right upper thorax	4 months	Pulmonary
Fischer G (1987)[6]	71/M	Back	4 weeks	Pulmonary
Cecchi R (1999)[7]	76/F	Right side of face	8 months	No involvement
Corazza *et al* (1999)[5]	70/F	Right upper thorax Right arm	2 months	No involvement
Requena (1998)[8]	51/F	Back	9 weeks	No involvement
Present report (2012)	50/M	Right scapular region	18 months	Pulmonary

drugs, chloroquine, isotretinoin, allopurinol, thalidomide and tetracyclines and carbon dioxide laser.[9]

Surveillance of the behavior of scars form an important part of examination in a patient with suspected or proven sarcoidosis, as biopsy of these scars is easily performed and alleviates the need of an invasive exploration to establish the diagnosis of sarcoidosis with systemic involvement. In the present report, diagnosis of post herpes zoster scar sarcoidosis helped confirm pulmonary involvement. In a patient with isolated cutaneous lesion/s, a careful and prolonged follow-up is strongly recommended due to the potential risk of developing systemic sarcoidosis.

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Cite this article as: Singal A, Vij A, Pandhi D. Post herpes-zoster scar sarcoidosis with pulmonary involvement. Indian Dermatol Online J 2014;5:77-9.

Source of Support: Nil, **Conflict of Interest:** None declared.