

Case Report

Multiple Yellowish Plaques and Nodules in a Young Man: A Case of Multiple Cutaneous Reticulohistiocytomas

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Abstract

Observations: Multicentric reticulohistiocytosis (MR) and multiple cutaneous reticulohistiocytomas (MCR) are exceedingly rare granulomatous conditions that belong to the reticulohistiocytosis spectrum of disorders. Reticulohistiocytoma usually presents as a firm, skin-colored, yellowish or reddish papule or nodule. We report here a case of multiple cutaneous reticulohistiocytomas in a young man for its rarity and to emphasize the importance of considering reticulohistiocytoma in the differential diagnoses of persistent yellowish plaques or nodules on skin.

Introduction

Multicentric reticulohistiocytosis (MR) and multiple cutaneous reticulohistiocytomas (MCR) are extremely rare idiopathic granulomatous conditions that belong to the reticulohistiocytosis spectrum of disorders [1].

Reticulohistiocytoma, the primary skin lesion for both the diseases, usually presents as a firm, skin-colored, yellowish, or reddish papule or nodule. In MR, extensive skin lesions occur in association with a severe, often destructive, arthropathy and other systemic features. In contrast, MCR is characterized by multiple cutaneous lesions, identical histologically to those seen in MR, developing in the absence of arthritis or other systemic lesions [2].

MCR is a very uncommon condition; a *PubMed* search revealed only 12 earlier reports in the English language literature. We report here a case of multiple cutaneous reticulohistiocytomas in a young man for its

rarity and to emphasize the importance of considering reticulohistiocytoma in the differential diagnoses of persistent yellowish plaques or nodules on skin.



Figure 1. Large plaque studded with multiple papules and nodules. A few satellite papules are seen around the lesion.



Figure 2. Multiple plaques over lateral aspect of the right thigh

Case Report

A 38-year-old man presented with a gradual onset of multiple asymptomatic swellings of variable sizes on his upper arms and right thigh for the preceding one and a half years. There was no history of local trauma, discharge, or joint pain. There were no constitutional symptoms and complaints referable to other systems. Cutane-

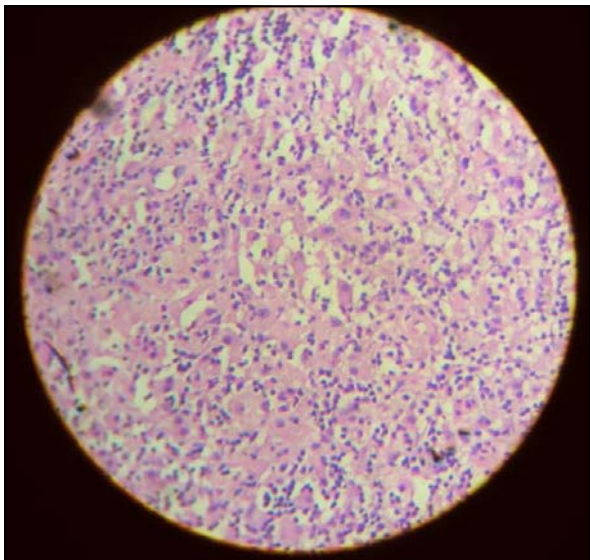


Figure 4. Dermal infiltrates composed of large histiocytes with abundant eosinophilic clear cytoplasm with a ground glass appearance and vesicular nuclei admixed with lymphocytes and plasma cells (H&E, original magnification x 400).

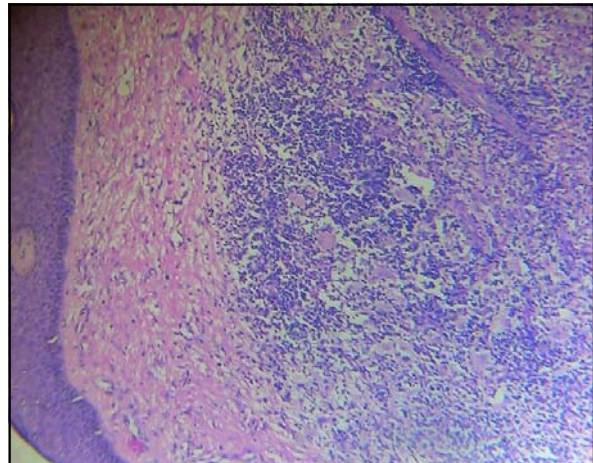


Figure 3. Flattening of epidermis, presence of a Grenz zone, intradermal cellular infiltrate containing large histiocytes having abundant eosinophilic clear cytoplasm with a ground glass appearance (H&E, original magnification x 100).

ous examination showed multiple yellowish-brown plaques on his left deltoid area (6 cm X 6 cm size) (**Figure 1**), medial aspect of the right upper arm (1cm X 1 cm in size), and lateral aspect of the right thigh (7cm X 5cm in size) (**Figure 2**). The plaques were soft to firm in consistency and studded with small papules and nodules giving rise to a lobulated, smooth and shiny appearance. In addition, multiple satellite papules of similar morphology were also seen around these plaques. The nails, mucosae, and hair were normal and systemic examination was non-contributory. Ultrasonography of abdomen, X-Ray chest and hands, complete hemogram, routine biochemical examinations, serum protein electrophoresis, and complete lipid profile were all within normal limits.

Lesional 5 mm punch biopsy showed flattening of epidermis, presence of a grenz zone (**Figure 3**), plenty of intra-dermal collection of cells composed of large histiocytes with abundant eosinophilic clear cytoplasm with a ground glass appearance, and vesicular nuclei. They were admixed with lymphocytes and plasma cells, neutrophils and eosinophils (**Figure 4**). Special staining for leprosy and mastocytosis were negative. Based on the clinical and histopathological findings a diagnosis of multiple reticulohistiocytomas was made. The patient was put on oral prednisolone (40 mg/day) therapy and showed significant response within two months before he was lost to follow up.

Discussion

Multiple cutaneous reticulohistiocytomas (reticulohistiocytic granulomas) represent a unique pattern in the spectrum of the reticulohistiocytoses, characterized by histiocytic proliferations of the skin and soft tis-

sues. It is believed that reticulohistiocytoses originates by proliferation and differentiation of an anomalous histiocytic clone in response to unknown stimuli [3]. Reticulohistiocytomas are usually solitary lesions of less than 1cm diameter [4]. However; multiple lesions and large reticulohistiocytomas have rarely been reported in the literature. Histopathology of reticulohistiocytoma often shows mid-dermal infiltration of mononuclear histiocytes and multinucleated histiocytes with a ground-glass appearance, and a variable number of vacuolated, spindle-shaped, and xanthomatized mononuclear histiocytes [5].

Histology, often supplemented by immunocytochemistry usually confirms the diagnosis of MCR. Immunohistochemical profiles show positivity with different macrophage markers including lysozyme, and alpha 1-antitrypsin. Vimentin is universally positive but all other markers like S100, desmin, and smooth muscle-specific actin, remain negative [5].

In our case, immunohistochemistry could not be performed due to local nonavailability and financial constraints. The main clinical as well as histological differential diagnosis of MCR is xanthogranuloma and some investigators consider these disorders

to be parts of the same nosologic spectrum of diseases [3]. Other diseases that need to be differentiated include sarcoidosis, xanthoma, mastocytosis, leprosy, and lymphoma. Systemic steroid with or without methotrexate usually improves the condition. Our patient also responded to systemic steroid. However, long-term vigilant follow-up is required in every case keeping in view the possibility of development of MR and other systemic associations of this rare entity.

References

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