CASE REPORT

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Prominent dermal accumulation of Russell bodies underlying pseudocarcinomatous hyperplasia with fungal infection

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ABSTRACT

Blockade of the secretion of immunoglobulins leads to their accumulation in plasma cells, resulting in condensed immunoglobulins in the rough endoplasmic reticulum of plasma cells, termed Russell bodies. They are sometimes found in lymphoplasmacellular inflammation of the intestinal mucosa and in lymphoid cell malignancies, but only very rarely in skin diseases. Here, we report an 86-year-old female who presented with a lesion with the prominent accumulation of Russell bodies underlying pseudocarcinomatous hyperplasia with fungal infection in the face. Immunohistochemical staining showed the cells containing Russell bodies to be positive for CD138 and the Russell bodies to be positive for immunoglobulin κ and λ light chains. The present case suggests that when inflammatory cell infiltration with abundant round intracellular eosinophilic materials is observed in the dermis, the dermal accumulation of Russell bodies should be considered in cases with reactive pseudocarcinomatous hyperplasia with fungal infection.

Keywords: fungus, immunoglobulin, plasma cell, Russell body

INTRODUCTION

When immunoglobulin secretion becomes blockaded, immunoglobulins can accumulate in plasma cells, resulting in condensed immunoglobulins in the rough endoplasmic reticulum of plasma cells. These are called Russell bodies, and while they are sometimes found in lymphoplasmacellular inflammation in the intestinal mucosa and lymphoid cell malignancies, they are only very rarely found in skin diseases.[1,2] Here, we report a lesion of pseudocarcinomatous hyperplasia with superficial fungal infection in the face, showing prominent intracellular and extracellular Russell bodies in the underlying dermis.

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CASE PRESENTATION

An 86-year-old woman presented with a 3-month history of scaly erythema on the face. Physical examination showed a reddish, irregularly shaped plaque with scales on the right cheek (Figure 1A). Potassium hydroxide examination of the scales revealed numerous hyphae. She had

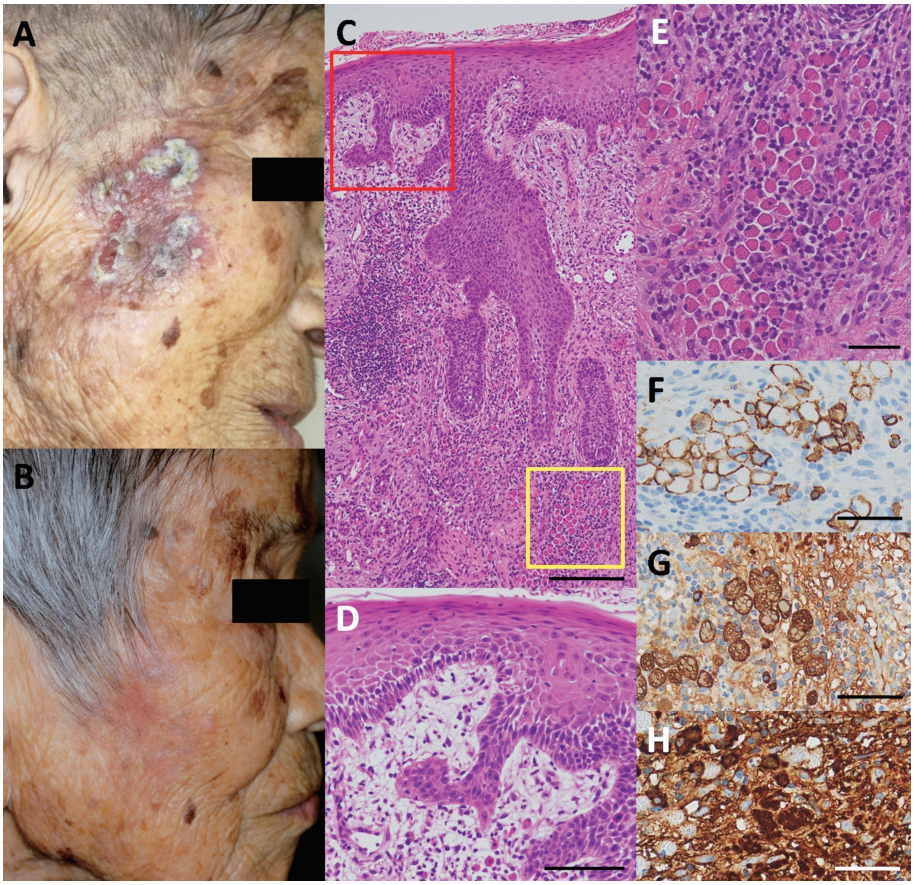

Fig. 1 The clinical appearance and histopathology of hyperkeratotic erythema on the patient's face

Fig. 1A: Scaling erythema is seen on the right cheek.

Fig. 1B: The hyperkeratotic erythema improved significantly after topical treatment with an antifungal agent.

Fig. 1C: Microscopy reveals acanthosis and hyperkeratosis with parakeratosis in the epidermis. Inflammatory cell infiltration with large numbers of round eosinophilic bodies is seen in the superficial and mid dermis. The areas bordered by the red rectangle and the yellow rectangle are enlarged in (D) and (E), respectively (hematoxylin and eosin staining, scale bar: 200 μm).

Fig. 1D: Pseudocarcinomatous hyperplasia is seen mainly in the basal layer (hematoxylin and eosin staining, scale bar: 100 μm).

Fig. 1E: Intracellular eosinophilic bodies are seen predominantly in the inflammatory cell infiltration in the dermis (hematoxylin and eosin staining, scale bar: 50 μm).

Fig. 1F: Cells containing the eosinophilic bodies are positive for anti-CD138 antibodies and are thought to be plasma cells (anti-CD138 staining, scale bar: 50 μm).

Fig. 1G–1H: The round eosinophilic bodies are positive for immunoglobulin κ light chains (G) and immunoglobulin λ light chains (H), indicating that the eosinophilic bodies are Russell bodies (G, anti-κ light chains; H, anti-λ light chains: scale bars: 50 μm).

been under hemodialysis due to renal failure. She had no history of blood disorders. A skin biopsy histopathologically showed irregular acanthosis, hyperkeratosis, and parakeratosis in the lesional epidermis, and basal cells in the epidermis showed enlarged, irregular, hyperchromatic nuclei (Figure 1C and D). There were no dermal granulomatous lesions such as those seen in Majocchi granulomas. From these findings, pseudocarcinomatous hyperplasia with superficial fungal infection was diagnosed. In addition, inflammatory cell infiltration, along with abundant round eosinophilic materials, was apparent in the dermis (Figure 1C and E). Immunohistochemical staining revealed the cells with cytoplasmic eosinophilic materials to be positive for CD138 (Figure 1F) and the round eosinophilic bodies to be positive for immunoglobulin κ and λ light chains (Figure 1G and H). From these findings, the prominent eosinophilic bodies in the underlying dermis were considered to be Russell bodies. The infiltrating cells with cytoplasmic eosinophilic bodies were considered to be polyclonal plasma cells with conspicuous Russell bodies. The plasma cells showed neither nuclear atypia nor mitosis. Blood tests ruled out syphilis and multiple myeloma. We treated the fungal infection with a topical antifungal agent, and the lesion improved significantly (Figure 1B).

DISCUSSION

Gastritis with the infiltration of Russell body-containing plasma cells in the gastric mucosa is known as Russell body gastritis.[3] A number of cases of Russell body gastritis associated with *Helicobacter pylori* infection have been reported.[4] A chronic bacterial infection can be the antigenic stimulus for the formation or accumulation of Russell bodies in the gastric mucosa.[4]

Concerning skin diseases, a case with actinic keratosis showing conspicuous Russell body accumulation in the dermis was reported.[5] In that case, no fungal infection was described in the actinic keratosis lesion.[5] In the present patient, the skin lesion was successfully treated with a topical antifungal agent, and the lesion largely disappeared in 3 months. Thus, we consider that actinic keratosis can be ruled out in the present case. In addition, a patient with chronic syphilis was reported to show dermal Russell body accumulation in a phimosis lesion.[2]

To our knowledge, the present patient is the first reported case of prominent dermal Russel body accumulation associated with pseudocarcinomatous hyperplasia with superficial fungal infection. In the present case, the patient had a facial lesion with the prominent accumulation of Russell bodies in the dermis beneath pseudocarcinomatous hyperplasia with superficial fungal infection. We speculate that the Russell bodies could have been produced by the chronic antigenic stimulus from the superficial fungal infection, similarly to the Russell body production in Russell body gastritis associated with *Helicobacter pylori* infection. We searched the literature for reports of patients with cutaneous superficial fungal infection and Russell body accumulation. However, no such reports were found. In addition, in the present skin lesion, no granulomatous lesions caused by the fungal infection were observed in the dermis. Thus, we were unable to obtain direct evidence of any association between the superficial fungal infection and the Russell body production. Furthermore, we speculate that inflammatory reactions to the pseudocarcinomatous hyperplasia in the lesional epidermis might be associated with the Russell body production. We have to accumulate more cases with cutaneous Russell body accumulation and to study their skin lesions in detail to elucidate the exact mechanisms of Russell body accumulation in the skin.

Eosinophilic bodies morphologically similar to Russell bodies include amyloids, colloid bodies, eosinophilic bodies seen in cryptococcosis and other fungal infections, and Michaelis–Gutmann bodies found in histiocytes in malakoplakia. These similar structures can be ruled out by specific stains.[5]

CONCLUSION

The present case suggests that when inflammatory cell infiltration with abundant round intracellular eosinophilic materials is observed in the dermis, the dermal accumulation of Russell bodies should be considered in cases with pseudocarcinomatous hyperplasia with superficial fungal infection. The further accumulation of similar cases is needed to elucidate the molecular mechanisms behind Russel body formation associated with pseudocarcinomatous hyperplasia with cutaneous superficial fungal infection.

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None.

CONFLICTS OF INTEREST

The authors have no conflicts of interest to declare.

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