Necrobiotic xanthogranuloma after penicillin injection: A case report

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Necrobiotic xanthogranuloma (NXG) is a rare, progressive, non-Langerhans cell histiocytosis characterized by multiple yellowish indurated plaques and subcutaneous nodules that frequently involve the face, trunk, and extremities. Extracutaneous manifestations and paraproteinemia are common.

Here, we report the case of a 47-year-old female who referred to the Dermatology Clinic of Razi Hospital, Tehran, Iran with complaints of a yellowish swelling on her right buttock after an intramuscular injection of penicillin. A similar lesion gradually developed on her supraorbital areas after several months. Histopathological findings included granulomatous inflammation of subdermal tissue with necrobiotic foci, which confirmed the diagnosis of NXG.

The patient was treated with an intralesional injection of triamcinolone and oral prednisolone. After 12 months of treatment, the plaques became considerably flatter and smaller.

Keywords: necrobiotic xanthogranuloma**,** injection, penicillin

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INTRODUCTION

Necrobiotic xanthogranuloma (NXG) is a rare, progressive, non-Langerhans cell histiocytosis characterized by multiple, yellowish indurated plaques and subcutaneous nodules that frequently involve the face, especially the periorbital region, trunk, and extremities [1,2].

Involvement of extracutaneous organs includes the ocular system, respiratory tract, liver, spleen, heart, lungs, kidneys, muscles, and central nervous system [1]. Abnormal laboratory results that include increased ESR, leukopenia, hypocomplementemia, and cryoglobulinemia are also common [1]. There is an associated paraproteinemia of monoclonal gammopathy usually of the IgG-κ type in 80% of patients along with an increased risk of hematological and lymphoproliferative malignancies [3].

Histopathologic survey shows infiltrates that contain macrophages, foamy cells, and inflammatory cells in the dermis and subcutaneous tissue, cholesterol cleft, and necrobiosis [4].

The pathogenesis of NXG is unknown but theories indicate a strong association with monoclonal gammopathies [5].

CASE PRESENTATION

A 47-year-old female referred to our Dermatology Clinic with complaints of a yellowish swelling on

her right buttock and bilateral supraorbital areas. At first she noticed an asymptomatic gradually enlarging yellowish swelling on her right buttock two weeks after an intramuscular injection of penicillin on her right buttock. The same lesions gradually affected her bilateral supraorbital areas over several months. On clinical examination, we detected a firm, non-tender, yellow-red, sharply demarked indurated plaque on her right buttock (Figure 1) and bilateral supraorbital areas (Figure 2).

The patient underwent two incisional biopsies from the buttock and supraorbital skin with a differential diagnoses of NXG, injection induced panniculitis, sarcoidosis, and lipogranuloma. The sections of the buttock skin lesion biopsy showed granulomatous inflammation of subdermal tissue and some dermal necrobiotic foci. The inflammation contained lymphohistiocytes, foamy macrophages, and giant cells (Figure 3). Sections of the supraorbital skin lesion biopsy showed multiple dermal foci of foamy macrophage infiltration, mostly around the dermal vessels (Figure 4). The histopathologic findings of the buttock skin lesion were compatible with NXG versus xanthoma in the supraorbital skin lesion. Some reports noted cases of NXG with no evidence of necrobiosis despite presenting other confirmatory pathologic features such as infiltrates of foamy macrophages and inflammatory cells in the dermis and subcutaneous tissue [6].

A diagnosis of NXG was based on characteristic clinical and histopathological findings, and the patient underwent investigations for associated disorders. Ophthalmological examination revealed no pathologic finding. General physical examination revealed no regional lymphadenopathy or hepatosplenomegaly. Laboratory investigations were normal for blood counts, fasting blood sugar, lipid profile, liver function tests, erythrocyte sedimentation rate (ESR), complement levels, LDH, serum β_2-microglobulin level, and peripheral

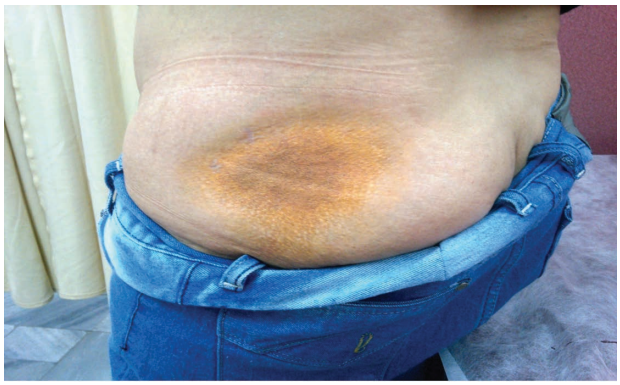

Figure 1. Indurated plaque on right buttock.

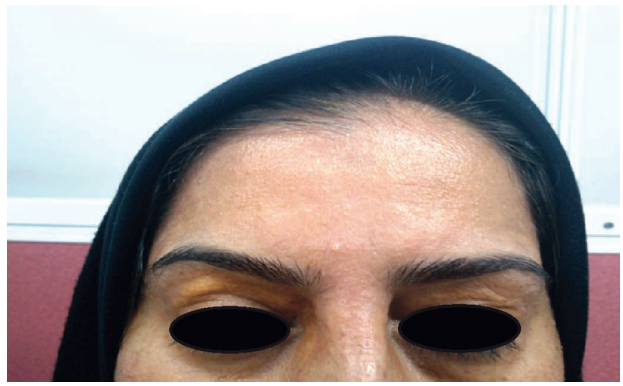

Figure 2. Bilateral supraorbital yellowish plaques.

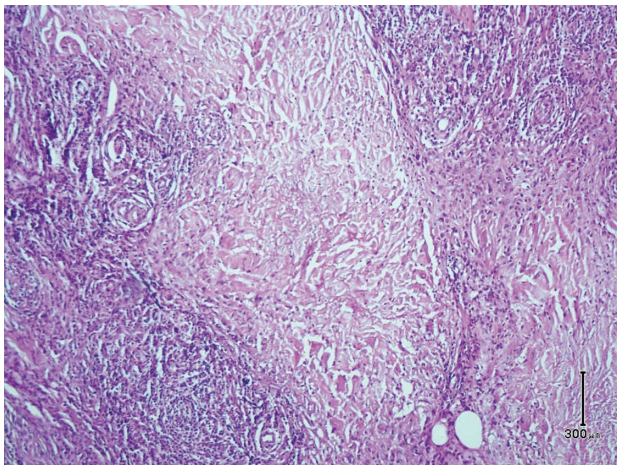

Figure 3. Histology of the buttock skin lesion biopsy showed granulomatous inflammation of subdermal tissue and some dermal necrobiotic foci (H & E, 100×).

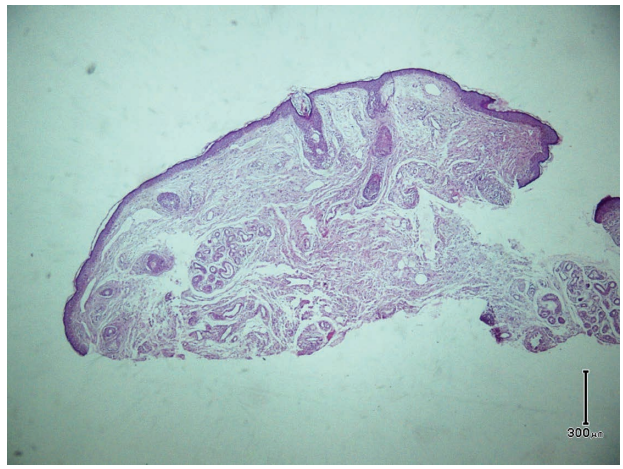

Figure 4. Histology of the supraorbital skin lesion biopsy showed multiple dermal foci of foamy macrophage infiltration, mostly around the dermal vessels (H & E, 40×).

blood smear. A chest x-ray, echocardiography, and ultrasound evaluation of the abdomen and pelvis showed no abnormal findings. Serum protein electrophoresis revealed polyclonal gammopathy of unknown significance.

DISCUSSION

The patient mentioned that the lesion on her right buttock started about two weeks after an intramuscular injection of penicillin at the same site; the periocular lesions appeared after the buttock lesion. The clinical features of NXG in this patient were similar to other cases of NXG without any predisposing factors. As reported by Ugurlu *et al.* [2], most NXG lesions initially occurred on the extremities or trunk and subsequently affected the periocular region.

Evidence of NXG at the site of a burn scar [7] has been reported, but we were unable to find any report that indicated a relationship between an injection or any other trauma and incidence of NXG in the literature review.

NXG may affect extracutaneous organs, including the ocular system, respiratory tract, liver, spleen, heart, lungs, kidneys, muscles, and central nervous system [1]. The current study patient had no pathologic findings on her medical history, physical examination, and paraclinical investigations indicated any systemic involvement.

Paraproteinemia is highly associated with NXG. A monoclonal gammopathy of IgG on serum protein electrophoresis in approximately 80% of patients has been reported [1-3]. However, cases of NXG with no paraproteinemia and significant laboratory abnormalities were reported [8]. In the current study patient, serum protein electrophoresis revealed only an undetermined significance of polyclonal gammopathy and no evidence of monoclonal gammopathy or hematologic malignancy per hematologic survey. Ugurlu *et al.* reported an interval between 8 years before the skin lesions appeared to 11 years after for incidence of associated malignancy based a survey of 26 patients with NXG [2]. Therefore, patients with NXG need life-long follow up to detect any accompanied malignancies.

Improvements in skin lesions have been reported in some patients treated with intralesional and systemic corticosteroids [9,10]. The current study patient received monthly intralesional injections of triamcinolone 8 mg/mL for the supraorbital lesion and 15 mg/mL for the buttock lesion. An oral prednisolone (30 mg daily) taper to 10 mg during 12 months was also prescribed. The plaques became flatter and smaller after 12 months of treatment.

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