■ Sun-475 ■

A case of hydroxychloquine-induced pigmentation on sun-exposed areas in a lupus patient

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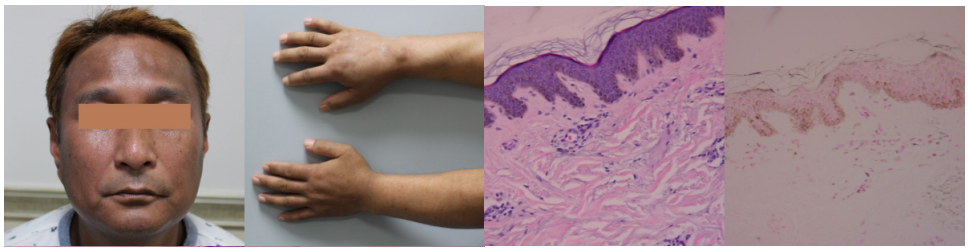
Antimalarial drug-induced skin pigmentation has been reported about 10 % of patients, but most cases are from the use of chlorquine, and cases from hydroxychloroquine (HCQ) are rare. Skin pigmentation predominates on the anterior side of the shins but can also be seen in the face, forearm, mouth mucosa and nail beds. Differential diagnosis includes melasma, Addision's disease, hemochromatosis, hyperthyroidism, and vitamin B12 deficiency. Heavy metal intake also can cause slate-gray (mercury, silver), blue- gray (bismuth, gold) or, gray in sun-exposed areas (silver sulfadiazine) or bronze (arsenic) pigmentation.The mechanism is poorly understood.Bruising, steroid use, oral anticoagulants, antiplatelet agents, and skin trauma is known to be the risk factors. We experienced a patient withlupus accompanied by pigmentation on his face and hands during the administration of HCQ, diagnosed as HCQ induced cutaneous pigmentation based upon clinical and histopathologic findings, and report here. **Case reports:** A 48-year old man was diagnosed as lupus based on findings including 2nd, 3rd proximal interphalangeal arthritis, photosensitivity, Raynaud phenomenon, positive ANA at 1;320 in a nucleolar pattern, positive anti-double stranded DNA and anti-cardiolipin antibody in March 2018. HCQ 400mg and prenisolone 5mg were prescribed, and after 3 months, blue-gray pigmentation was apparent on his face and both hands, which are supposed to be sun-exposed areas. There was no involvement of the shin, hard palate, mucous membrane or nails. Serum levels of T3, free T4, TSH, cortisol, ACTH and prolactin were all within normal limits. Skin biopsy from the face, hand showed coarse granular pigment deposition interstitially. S-100 protein staining for melanin and Perls Prussian blue staining for iron were positive. We considered this case as HCQ-induced pigmentation based on the temporal relationship between pigmentation and onset of drug effects, and exclusion of other causes.



■ Sun-476 ■

A rare case of diffuse large B-Cell lymphoma developing in a patient with IgG4-related disease

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Introduction: IgG4-related disease (IgG4-RD) is a fibro-inflammatory entity characterized by IgG4-positive lymphoplasmacytic infiltration in multiple organs including salivary glands, orbit and retroperitoneum. Lymphomas that occur in the background of IgG4-RD are uncommon although the association of lymphoma and autoimmune diseases such as rheumatoid arthritis and Sjögren's syndrome is frequently reported. Especially, asynchronous occurrence of diffuse large B cell lymphoma (DLBCL) and IgG4-RD is extremely rare. Here, we report on a case of DLBCL in a patient with longstanding IgG4-RD. **Case:** A 51-year-old male visited our clinic with a complaint of multiple painless masses on right submandibular area and both shoulders for the past 3 months. Laboratory findings showed elevated serum level of IgG to 4867.2 mg/dl (normal range, 700-1600 mg/dl). We performed excisional biopsy of right submandibular mass. Histopathological findings showed infiltration of plasma cells, eosinophils and histiocytes with strongly positive IgG4 on immune-histochemical staining. Pathologic diagnosis was IgG4-RD. After he was started on steroid therapy, the size of the masses decreased dramatically. Since then, he remained stable for 6 years taking low dose steroid and azathioprine. After 6 years of diagnosis, multiple masses were newly developed on both anterior thigh areas and the mass size on both shoulders suddenly increased (Figure 1). On Computed tomography, a subcutaneous fatty layer and intramuscular spaces of both thighs, buttocks, back and chest wall masses were seen with neck lymphadenopathy (Figure 2). Laboratory findings showed elevated serum level of lactate dehydrogenase, beta-2 microglobulin with normal Serum IgG level. Excisional biopsy of soft tissue mass on anterior thigh area revealed DLBCL with positive staining for pan-B cell markers. **Conclusion:** Clinicians should be aware of the potential for the development of lymphoma in patients with IgG4-RD when new extranodal masses occur or extranodal masses persist despite appropriate therapy for IgG4-RD.