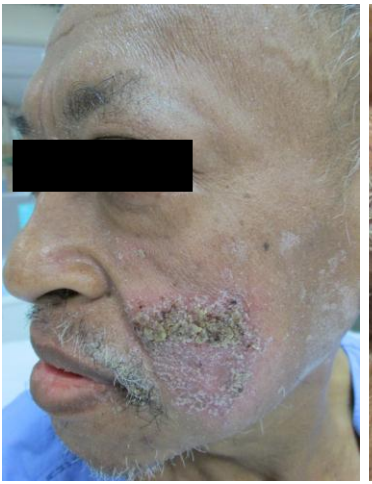
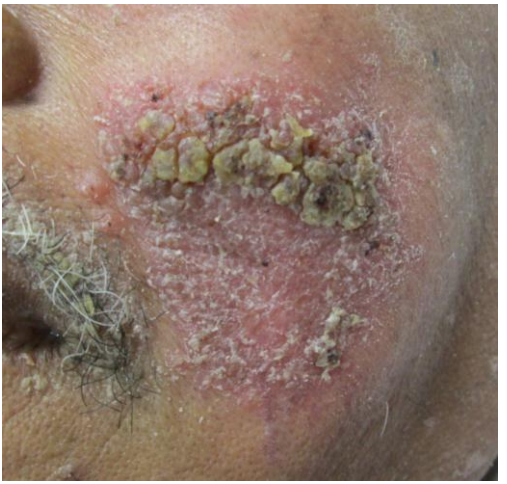
Case 5

A 61-year-old Thai man from Bangkok.

Chief complaint: Erythematous, verrucous plaque at face for 7 months.





Present illness:

Seven months earlier, the patient noticed small solitary erythematous non-tender nodule on left cheek after he was bitten by unidentified fly while he was working in Saudi Arabia. The lesion was aspirated for pus by physician in Saudi Arabia, without other topical or systemic treatment.

The patient returned to Thailand 6 months earlier. His lesion was treated with topical fusidic acid ointment. However, the lesion still expanded to a large, non-tender, verrucous plaque.

He denied symptom of weight loss or prolonged fever.

Personal history:

He was a labor worker in Saudi Arabia for 20 years

He drank 1 bottle of rice whisky/day for 20 years

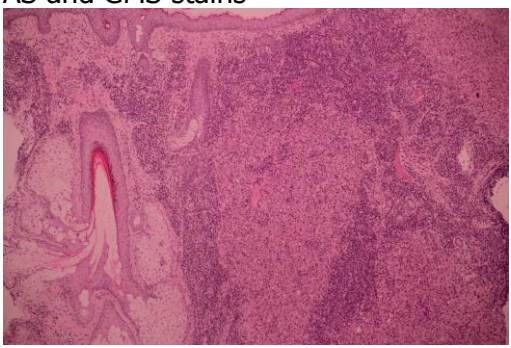
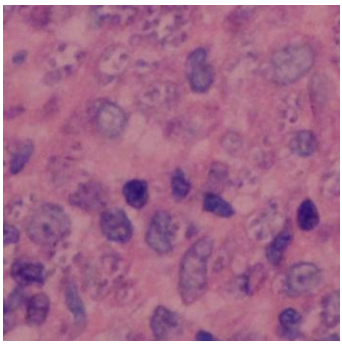
Past history

Alcoholic cirrhosis, esophageal varices, gastric and duodenal ulcer

Physical examination
A Thai man, good consciousness, afebrile.
HEENT: mild pale conjunctivae, no icteric sclera
Lymph node: no palpable axillary, supraclavicular and inguinal lymph node.
Lungs: clear
CVS: normal S1 and S2 , no murmur
Abdomen: soft, not tender, no hepatosplenomegaly
Extremities: no edema
Neurological: grossly intact

Skin examination
Solitary, well-defined, slightly elevated, erythematous, verrucous surface plaque at left cheek.

Histopathology (S13-15645, left cheek)
• Dense diffuse inflammatory-cell infiltrate of epithelioid histiocytes, surrounded by lymphocytes and plasma cells in the entire dermis.
• Cytoplasm of histiocytes filled with blue-gray round to oval (Donovan) bodies which are positive with Giemsa but negative with PAS and GMS stains

Investigation:
CBC: Hb 7.4 g/dL, Hct 21.9%, Plt 237,000/uL
WBC 5,200 (N69%, L21%, M9%, E1%)

LFT: AST/ALT 46/30 ALP/GGT 83/55
TB/DB 1.7/0.7 TP/Alb 80/28
Anti HIV: negative
Tissue PCR: positive for *Leishmania major*

Diagnosis: Localized cutaneous leishmaniasis
Treatment: Fluconazole (200mg) 1 tablet oral daily
Cryotherapy weekly
Presenter: Ruedee Phasukthaworn, M.D.
Consultant: Natta Rajatanavin, M.D.

Discussion

Our patient presented with slow progressive verrucous plaque at left cheek, after the bite of unidentified fly in Saudi Arabia. Our differential diagnoses are chronic infection (leishmaniasis, tuberculosis verrucosa cutis, chromoblastomycosis and NTM infection), inflammation (impetiginized seborrheic dermatitis) and malignancy (Bowen's disase, verrucous carcinoma). We performed skin biopsy for histopathology, PCR for leishmania spp., aerobe, TB and fungal culture. The result showed numerous intracellular amastigotes containing macrophages at upper dermis from histopathology. The diagnosis of cutaneous leishmaniasis was confirmed by positive PCR for *Leishmania major*.

The leishmaniasis are group of diseases caused by several species of the genus *Leishmania*. Transmission is via the bite of infected female sandflies from the genera *Phlebotomus* and *Lutzomyia*. In general, canine and rodent species are reservoirs for *Leishmania*, humans are accidental hosts. The prevalence of the disease is in excess of 12 million cases in 88 countries which 90% occur in only 7 countries: Afghanistan, Algeria, Brazil, Iran, Peru, Saudi Arabia, and Syria [1].

Clinical presentations of Leishmanisis have wide spectrum, depend on species of *Leishmania*, number of inoculated parasites

and host immune response [2,3]. The 4 clinical patterns of the disease in the human host are localized cutaneous leishmaniasis(LCL), diffuse cutaneous leishmaniasis(DCL), mucocutaneous leishmaniasis(MCL), and visceral leishmaniasis(VL). Acute LCL is defined by spontaneous regression within 1 year. LCL is generally categorized into two groups; old world and new world LCL.

Old world LCL mainly locates in Middle East Asia, Central Asia, India, Pakistan, East Africa and Eastern Mediterranean. The causative agents are *L. major, L. tropica, L. aethiopica*, and rarely *L. infantum*. Clinical presentations are divided into two types, as described in table 1. However other cutaneous lesions have been reported such as; eczematoid, psoriasiform, zosteriod, erysipeloid, annular, palmoplantar and keloidal lesions.

Table 1

Localized cutaneous leishmaniasis in old world		
Type	Moist type	Dry type
Causative species	*L. major*	*L. tropica*
Clinical	Multiple insect-bite like lesion on exposed area, often become ulcer	Solitary lesion , common on face Infrequently become ulcer
Healing time	6 months	12 months
Outcome	Self-healing	Risk for developing into chronic LCL

New world LCL is mainly caused by y *L. mexicana* and *L. braziliensis*, which are found in central and south America. Cutaneous lesions are varied and non specific.

Our patient developed cutaneous leishmaniasis in Saudi Arabia, which locates in old world distribution. The lesion is solitary slow progressive plaque on left cheek, no ulcerative lesion, which is compatible *for L. tropica* infection. However, clinical manifestation is inadequate to make the final diagnosis. PCR from skin biopsy of our patient reveals *L. major*.

Diagnostic of acute LCL confirmed by histopathology, which shows dense lymphohistiocytic dermal infiltration with presence of amastigotes in dermal histiocyte. Culture from skin biopsy or aspirested-fluid in special media sush as Novy-Mac-Neal-Nicolle (NNN) or chick embryo medium, remains gold standard with 50% sensitivity. PCR from tissue is useful method for identify *Leishmania* with higher sensitivity and specificity [4].

Systemic treatments are required in DLD, MCL and VL. LCL is naturally self-healing disease, however treatment accelerates healing, prevents disfiguring scar and disseminated disease. Multiple or large lesions, persistence for > 6 months and location over a joint are indicated for therapy [5]. Antimony derivatives; Sodium stibogluconate and meglumine are standard treatment for cutaneous leishmaniasis. Alternative treatments for CL are avialable in both systemic and local therapy such as; itraconazole[6], ketoconazole [7, 8], fluconazole[9, 10], zinc sulfate[11,12], topical paromomycin[13,14] and cryotherapy[15].

The treatment with daily oral fluconazole 200 mg and weekly cryotherapy was recently commenced in our patient. The long term clinical response has to be followed.

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