CASE REPORT

THE FIRST REPORTED CASE OF AUTOCHTHONOUS CUTANEOUS LEISHMANIASIS IN THAILAND

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Abstract. Thailand is not an endemic area for leishmaniasis. Several cases of autochthonous visceral leishmaniasis have been reported from Thailand but cutaneous leishmaniasis has never been reported. We reported a three-year-old girl who presented with a chronic ulcer on her cheek which proved to be cutaneous leishmaniasis. The diagnosis was made by finding amastigotes on skin biopsy; the patient had a therapeutic response to itraconazole.

Keyword: cutaneous leishmaniasis, *Phlebotomus*, Thailand

INTRODUCTION

Leishmaniasis, caused by *Leishmania* spp, is an obligated intracellular dimorphic protozoan transmitted in promastigote form by the *Phlebotomus* sand fly. Leishmania exists in the amastigote form in the human reticuloendothelial system.

Thailand is not endemic for leishmaniasis but there have been several reported cases of imported visceral and cutaneous leismaniasis since 1960 (Laohapaibol and Siampakdi, 1960; Viriyavejakul *et al*, 1997) The first reported case of autochthonous leishmaniasis was from Southern Thailand (Thisyakorn *et al*, 1999). Since then, several other cases have been reported from Thailand (Kongkaew *et al*, 2007; Maharom *et al*, 2008; Sukmee *et al*, 2008). All the autochthonous reported cases have been visceral leishmaniasis and no potential vector was identified.

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CASE REPORT

We report a three-year-old girl with cutaneous leishmaniasis. This is the first reported case of autochthonous cutaneous leishmaniasis from Thailand. The patient is from Lop Buri Province, central Thailand. The girl was referred to the Institute of Dermatology, Department of Medical Services, Ministry of Public Health, Bangkok, Thailand with a chronic asymptomatic ulcer on her left cheek for one month. Her mother noticed the lesion appeared

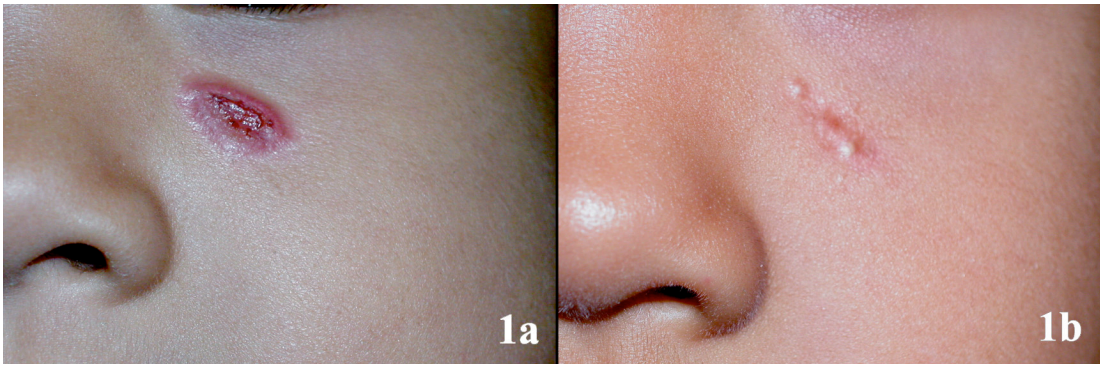

Fig 1–A three-year-old girl with a chronic ulcer on her left cheek before treatment (1a) and at 7-month follow-up after treatment with oral itraconazole (1b).

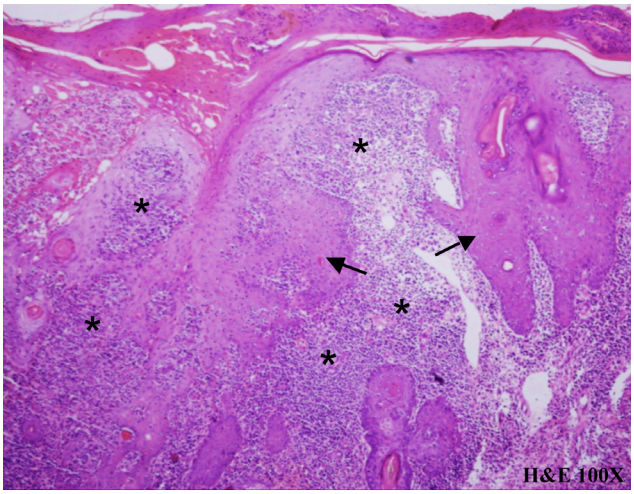

Fig 2–Histopathology of the ulcer showing pseudoepitheliomatous hyperplasia of the epidermis (arrows) and dense infiltration of inflammatory cells in the dermis (asterisks); H&E stain; original magnification 100x.

as an erythematous papule on her left cheek that slowly progressed to an erythematous plaque and finally to a crusted ulcer. She was treated at a local clinic with a combination of a topical antibacterial ointment and oral dicloxacillin but the lesion did not improve. She was otherwise healthy and never had traveled abroad. Her house was in a military village and she lived with her parents and her sister; they had no pets. There were no reports of leishmaniasis from her village.

On physical examination, she had a solitary painless, dry, crusted ulcer with a raised erythematous border on her left cheek (Fig 1). She had no fever on examination and the rest of her vital signs and physical examination were within normal limits. A skin biopsy was done which revealed pseudoepitheliomatous hyperplasia of the epidermis and dense infiltration of inflammatory cells composed of histiocytes, lymphocytes and neutrophils in the upper dermis (Fig 2). There were numerous amastigotes within the histiocytes and extracellularly (Fig 3). A Periodic Acid Schiff's (PAS) stain showed distinct kinetoplasts in the parasites (Fig 4). The kinetoplasts were best seen with a smear sample. Bacterial, mycobacterial and fungal cultures were negative. A biphasic medium culture was not available. The tissue biopsy was not adequate for polymerase chain reaction study and her mother did not approve of another tissue

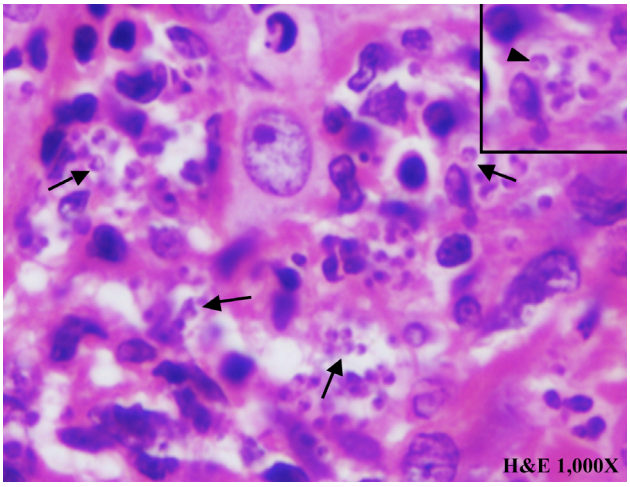

Fig 3–Higher magnification of the dermis showing numerous amastigotes within histiocytes (arrows). Kinetoplasts are discernible (inset, arrowhead) on H&E stain; original magnification 1,000x.

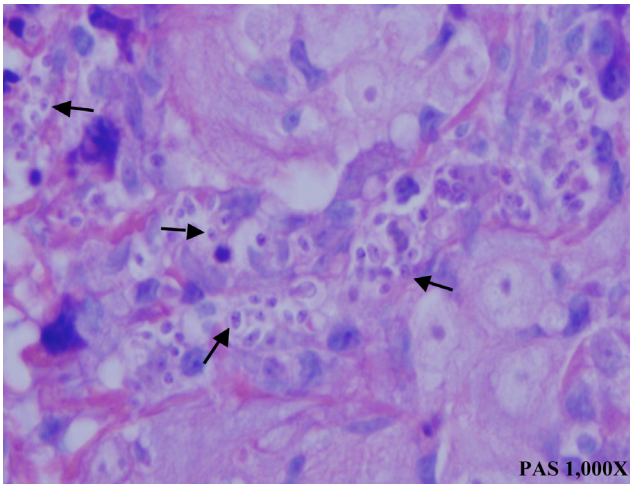

Fig 4–A PAS stain showing numerous amastigotes within histiocytes, each with a distinct kinetoplast (arrows) (original magnification 1,000x).

biopsy. Her complete blood count, liver function tests and abdominal ultrasound were unremarkable. She was diagnosed as having cutaneous leishmaniasis and treated with itraconazole 5 mg/kg/day for two months. The lesion subsided and did not recur by a 7-month follow-up visit (Fig 1b).

DISCUSSION

There have been several cases of autochthonous transmission of visceral leishmaniasis reported in Thailand but the vector of the disease was not identified in any of the cases (Thisyakorn *et al*, 1999, Kongkaew *et al*, 2007, Maharom *et al*, 2008, Sukmee *et al*, 2008). Since Thailand is non-endemic for leishmaniasis, there may have been other undiagnosed cases. In a survey of a cave in Saraburi Province (central Thailand), sandflies of the genera *Phlebotomus* and *Sergentomyia* were identified, which are known vectors of leishmaniasis (Polseela *et al*, 2007). The distribution of leishmaniasis had expanded in South and Southeast Asia (Katakura, 2009). Cattle, buffalos and other animals can be infected and become reservoir hosts. Cutaneous leishmaniais has several clinical presentations. It can be subclinical or causes papules, nodules, ulcerative lesions, atrophic scars, satellite lesions or sporotrichoid-like lesions. The incubation period averages several weeks. Our patient had a chronic ulcer which did not respond to antibacterial treatment. She was immunocompetent and had never been abroad. Cutaneous leishmaniasis was diagnosed on clinical and histopathological findings and the response to itraconazole. Identification of leishmania kinetoplasts in the histological section was the key to diagnosis in this patient.

There is no standard treatment for cutaneous leishmaniasis. Spontaneous remission can occur. Treatments include intravenous or intramuscular antimony therapy, oral ketoconazole, itraconazole, dapsone, paromomycin (intralesional, intravenous or topical), cryotherapy and surgical removal (Urcuyo and Zaias, 1982; Herwaldt, 1999). Recent studies have shown an ankylphosphocholine-miltefosine (an anti-cancer drug) to be an effective anti-leishmanial drug. Ongoing drug development for cutaneous leishmaniasis includes new compounds: 2-substituted quinoline alkaloids, and 8-aminoquinolines, which have been proven to be effective in animal studies (Seifert, 2011). This patient responded to itraconazole, which has been reported to be effective in the treatment of cutaneous leishmaniasis (Al-Fouzan *et al*, 1991).

Leishmaniasis is of public health concern since there have been several reported cases from various areas of Thailand. The incidence may be underestimated due to a lack of awareness and low prevalence. Surveying of vectors and reservoirs in the neighborhood of this patient is needed to document geographical distribution.

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