

INTRODUCED LEISHMANIASIS ON TAIWAN

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INTRODUCTION

We recently reported two cases of autochthonous cutaneous-subcutaneous leishmaniasis on Taiwan (Cross *et al.*, 1985). The patients were Taiwan aborigines who lived in their remote village since birth. No other cases of leishmaniasis were found in humans and animals in the village and it was speculated that the patients acquired their infections by mechanical arthropod transmission. Arthropods could have fed on a soldier from the China mainland stationed in the village and who may have had dermal leishmaniasis. The arthropod could have carried the parasite on the mouth parts and the organisms subsequently introduced into the patients. More than 100 cases of visceral leishmaniasis and post-kala-azar dermal leishmaniasis have been seen in mainland Chinese who migrated to Taiwan after World War II (Cheng, 1954; Cross *et al.*, 1985). Five of these imported cases occurred in former Chinese soldiers who were evacuated to Taiwan after 1949 and were seen and treated at the U.S. Naval Medical Research Unit No. 2 in Taipei, Taiwan. Two soldiers had visceral leishma-

niasis and three post-kala-azar dermal leishmaniasis.

CASE REPORTS

Case 1: The patient was a 31-year-old soldier from Kiangsu Province, China who arrived in Taiwan in 1949. He reported a past history of typhoid and malaria and questionable splenomegaly. In 1951 the skin over his lower lip became reddish, dry and fissured. Macular lesions gradually developed around his mouth, and over his face (Fig. 1, 2, 3), extremities (Fig. 4) and trunk. He was seen by several physicians during the next two years who suspected leprosy but could not confirm it.

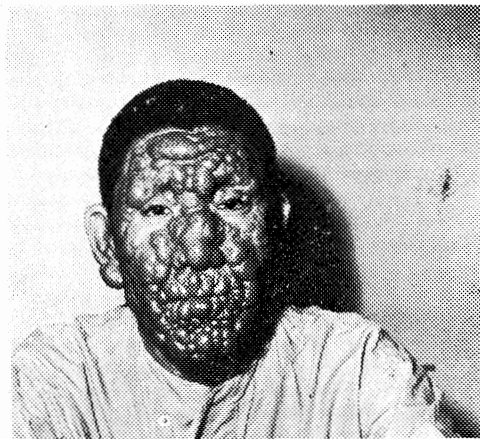


Fig. 1—Post-kala-azar dermal leishmanoid in a retired Chinese soldier who was first thought to have leprosy and hospitalized for 4 years. He was treated successfully with intravenous amphotericin B. Details of the case have been published elsewhere (Morgan *et al.*, 1962).

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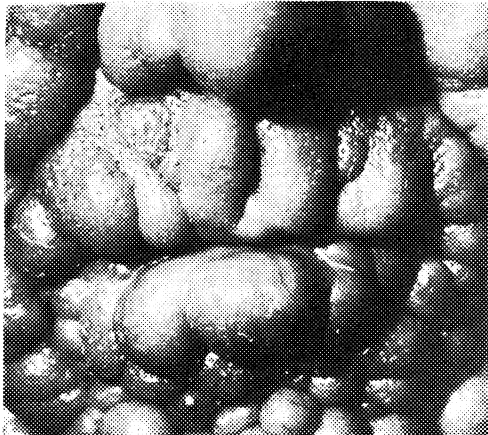


Fig. 2—Close-up photograph (Fig. 1) of the mouth of the patient with post-kala-azar dermal leishmaniasis showing nodules.

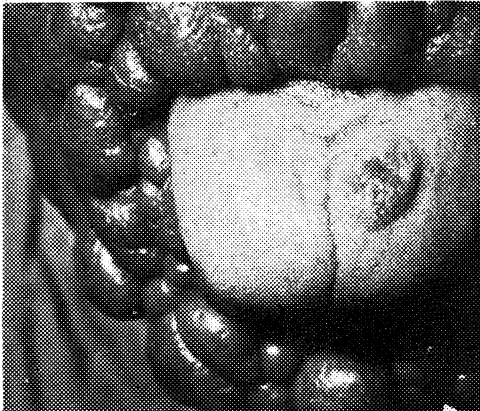


Fig. 3—Patient with post-kala-azar dermal leishmaniasis (Fig. 1) showing lesion on his lips and tongue.

He was admitted to a leprosarium in 1954 and in 1958 was sent to NAMRU-2 as a diagnostic problem. A biopsy was taken from a nodule and Leishman-Donovan bodies were found. Later, leishmanial organisms also were found in bone marrow. Ethylstibamine, atabrine and oral amphotericin B had no therapeutic effect. Hydroxystibamide caused clearance of some lesions but new ones developed even after two courses of the drug. Intravenous amphotericin B, however, resulted in complete cure (Fig. 5). The patient

became an employee of NAMRU-2 and never experienced relapse. Details of this case are reported elsewhere (Morgan *et al.*, 1962).

Case 2: A 42-year old male from Fukien Province, China was admitted to NAMRU-2 ward in April 1958 with a 3-year history of hepatosplenomegaly, fullness of the abdomen and general malaise. He had been in the Chinese army and arrived on Taiwan in 1951. He had a history of fever and chills for 3 months in 1951 and in 1953 he lost weight, had general malaise and hemoptysis. Abdominal enlargement, diminished urine and pitting edema developed in 1955. When seen in 1958 he was chronically ill with bronchial vesicular breathing, visible subcutaneous bleeding on the anterior chest wall, and a distended abdomen apparently with fluid. The liver was enlarged, soft, smooth and tender, and the spleen was described as being



Fig. 4—Legs of patient in Fig. 1 showing leishmanial lesions.



Fig. 5—Patient in Fig. 1 showing healing lesions following treatment with amphotericin B.

elastic and firm. The lower extremities revealed macular spots and pitting edema. The diagnosis of kala-azar was made by finding leishmanial organisms in liver and spleen biopsies. He was treated with two courses of ethylstibamine and was cured. He also became an employee of NAMRU-2 and was followed for several years.

Case 3: This 40-year-old from Kiansu Province had splenomegaly and was treated for kala-azar with antimony in 1939. In 1956 a small reddish papule developed on the neck and spread to the face, left ear, and both elbows. Similar nodules appeared on the scrotum in 1958, and in 1963 nodules appeared on the right ear and submandibular areas. There was no pain, itching or sensory disturbance. He was treated for leprosy and subsequently referred to NAMRU-2 in 1965.

At that time granulomatous erythematous lesions were present on the face, neck, ear, elbow and scrotum and the metatarsal joint of a right toe. Blebs were found in the mouth and lower lip (Fig. 6). The skin lesions were not itchy or painful. There were no palpable lymph nodes and the liver and spleen were normal. Bone marrow biopsy was negative, but leishmanial organisms were found in skin biopsies of the elbow and neck. The patient received 3 courses of sodium stibogluconate and the lesion completely disappeared after the third course.

Case 4: In 1944 this patient from Kiangsu Province had been treated for kala-azar on the China mainland. A skin lesion appeared on his neck in 1954 and during the next 12 years spread over most of his body. When seen at NAMRU-2 in 1967 the patient was 42 years of age and had a generalized skin condition characterized by large raised confluent erythematous lesions particularly on the trunk and face. Similar lesions were on the arms and legs but with an overlying scaly dermatitis (Fig. 7, 8). Leishmanial organisms were found in biopsied skin lesions by direct smear, culture and in histological preparations. The patient was treated with hydroxystilbamidine but refused continuation of treatment because of side effects. The parasite was still present in biopsied tissue taken when treatment stopped, but the organisms were fewer in number and they failed to become established in culture.

Case 5: This patient was 49 years of age when admitted to the NAMRU-2 ward with hepatosplenomegaly, anemia, leukopenia, hyperglobulinemia and axillary lymphnode enlargement. Leishman-Donovan bodies were found in bone marrow and splenic biopsy. He was born in Canton, China and traveled to many provinces of China after joining the Chinese army in 1939. He denied any illness except what he called malaria in 1949. He had a duodenal ulcer in 1963 and was told that he had

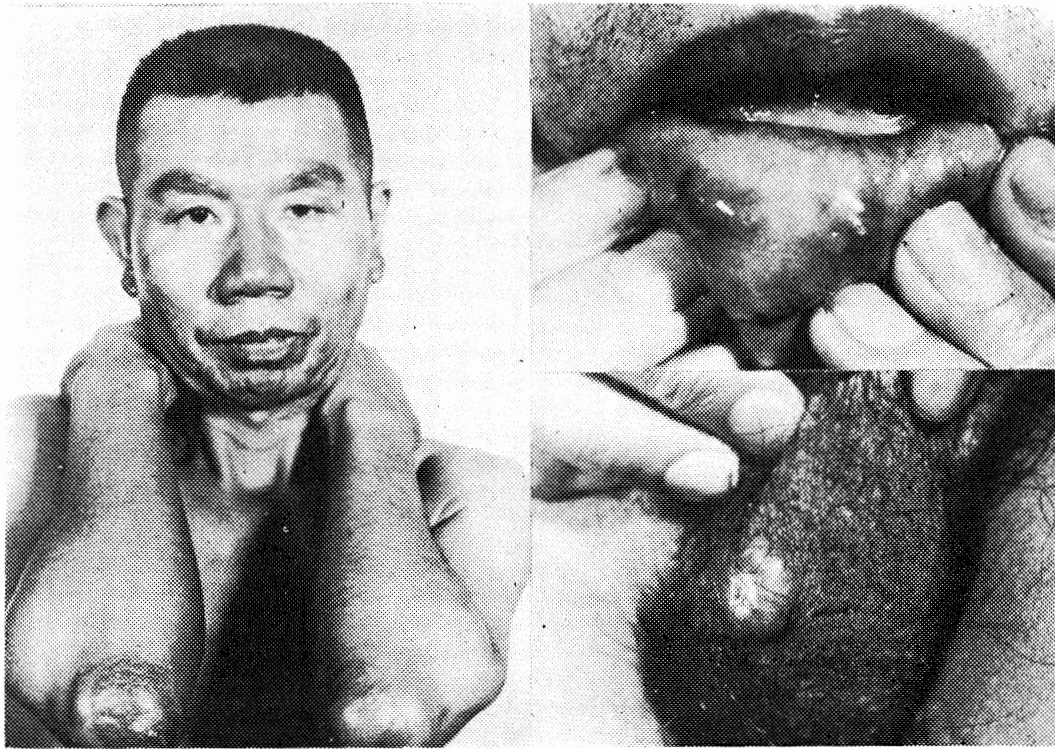


Fig. 6—Retired Chinese soldier who was treated for kala-azar on the China mainland. Seventeen years later he developed cutaneous lesions on his chin, lips, ears, elbows, and scrotum. Amastigotes were recovered from biopsied lesions.

splenomegaly in 1964. He was seen in the Chinese Veterans Hospital in 1967 and was subsequently referred to NAMRU-2. He was treated successfully with sodium stibogluconate and observed for several months. He failed to return for a follow-up bone marrow examination, however.

DISCUSSION

Prior to World War II leishmaniasis was not seen in any person on Taiwan. However, in 1942 Japanese soldiers that returned to Taiwan from the China mainland became symptomatic (Hongyo, 1942). Following World War II, over two million people from the China mainland were evacuated to Taiwan and following this massive immigration

leishmaniasis began to be reported. By 1954, 57 cases were reported among Chinese military personnel and their dependents and between 1948 and 1958 additional cases had been reported. Five of these patients were later referred to NAMRU-2 for study, treatment and follow up. The two indigenous cases of cutaneous-subcutaneous leishmaniasis were seen in 1968 and 1970. Except for two other cases of visceral leishmaniasis seen at other medical centers on Taiwan, no additional indigenous or imported cases of the disease had been seen and documented since 1970. Extensive studies were done by NAMRU-2 to determine the spread of the disease on the island but no evidence of the disease was found in humans and other mammals. Studies on the vectors of leish-



Fig. 7—This retired Chinese soldier was treated for kala-azar in China in 1944, began to develop skin lesions in 1954 and was treated for post-kala-azar dermal leishmaniasis in 1967. He had extensive lesions over his face, legs and trunk. This photograph shows lesions found on his back, arm, chest and abdomen.



Fig. 8—Same patient in Fig. 7 showing leishmaniasis on arm and abdomen.

maniasis, *Phlebotomus* sp. were done and 8 species have been identified. None were found infected with promastigotes, however. Only one of these species was suspected as feeding on human.

This report indicates that although an infectious disease may be imported into an area, it does not always become established and endemic to the new area. Had biological vectors (Cates and Lien, 1970), such as human feeding *Phlebotomus* sp., been indigenous to Taiwan the disease could have possibly become established.

SUMMARY

Leishmaniasis is not known to be indigenous to Taiwan but a number of imported

cases of visceral as well as post-kala-azar dermal leishmaniasis have been seen. Only two autochthonous cases of cutaneous-subcutaneous diseases have been documented in aborigines but no cases of visceral leishmaniasis have been reported. Although a significant number of imported cases of leishmaniasis have been seen, the disease has apparently not been established on the island.

REFERENCES

- CATES, M.D. and LIEN, J.C., (1970). The *Phlebotomus* of Taiwan. *J. Med. Ent.*, 7 : 529.
- CHENG, H.C., (1954). Exogenous kala-azar in Taiwan. *Chinese Med. J.*, 1 : 87.
- CROSS, J.H., GUNNING, J-J., DRUTZ, D.J. and LIEN, J.C., (1985). Autochthonous

- cutaneous-subcutaneous leishmaniasis on Taiwan. *Amer. J. Trop. Med. Hyg.*, 34 : 254.
- HONGYO, R., (1942). Some observations on blood and urine of kala-azar, especially on Tomita's urine reaction. *Taiwan Igakkai Zossi*, 41 : 279.
- MORGAN, F.M., WATTEN, R.H. and KUNTZ, R.E., (1962). Post-kala-azar dermal leishmaniasis - A case report from Taiwan (Formosa). *J. Formosan Med. Assoc.*, 61 : 282.