CASE REPORT

Imported gnathostomiasis manifesting as cutaneous larva migrans and Löffler's syndrome

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SUMMARY

Here, we report an unusual case of invasive gnathostomiasis in a returning traveller, with a shifting pattern of relapsing cutaneous disease. The previously fit and well 32-year-old man first presented with serpiginous, pruriginous erythematous tracks characteristic of cutaneous larva migrans shortly after returning from South-East Asia. He was systemically well with no other symptoms. After ivermectin therapy, he re-presented with respiratory symptoms, peripheral eosinophilia and transient pulmonary infiltrates; the classic triad of Löffler's syndrome associated with invasive helminth infection. *Gnathostoma spinigerum* immunoblot was positive. After a second round of ivermectin therapy his respiratory symptoms resolved, but the patient's cutaneous disease relapsed repeatedly over months, with migratory erythematous swellings appearing and settling after a few days. He was treated with a 21-day course of albendazole and is lesion free at 40 weeks post initial presentation.

BACKGROUND

Parasitic worms (helminths) cause significant morbidity globally, particularly among the poorest billion people on Earth.^{1 2} Helminth infections are also imported into non-endemic countries through increasing international travel. Ehrhardt and Burchard³ estimate that up to 5% of asymptomatic travellers returning from the tropics have eosinophilia, and 14%–48% of those suffer from parasitic infections such as schistosomiasis, filariasis or strongyloidiasis.

Helminths cause both cutaneous and visceral disease, and vary in pathogenicity from non-invasive, self-limiting manifestations like cutaneous larva migrans (CLM) to serious invasive forms capable of penetrating visceral organs and the central nervous system (CNS). This case highlights the multifarious forms of cutaneous gnathostomiasis, including mimicking CLM, the potential for *Gnathostoma* larvae to cause invasive visceral disease associated with peripheral eosinophilia and the difficulty in achieving long-term cure in human gnathostomiasis.

CASE PRESENTATION

A 32-year-old man was referred to the Ambulatory Care Unit (ACU) at Royal Sussex County Hospital in Brighton by his general practitioner (GP) with a rash on his left foot that appeared while he was on holiday in South-East Asia. The rash (figure 1)

consisted of two discrete lesions; both were sharply delineated, serpiginous erythematous tracks which were intensely itchy, a typical presentation for the creeping eruption of CLM. At first presentation, he had no other symptoms and was systemically well. He had no significant medical or family history and was normally fit and well, taking no regular medications. Clinical examination was unremarkable other than the rash.

Reviewing his travel history, he left the UK in September (outbound flight taken as day 1), travelling mostly within mainland Cambodia. From around day 13 he spent a week on the island of Koh Rong, where he reported running barefoot along the beach and having contact with stray dogs. He denied skin contact with fresh water. He drank from bottled water only, ate fresh fruit drinks and cooked meats including chicken and seafood. He noticed a generalised erythematous rash on his left foot on the final day in Koh Rong before returning to the UK via Kuala Lumpur on day 22. During his return stay-over in Kuala Lumpur, he developed a transient diarrhoeal illness with fever, sweats and loose watery stools that lasted about 2 days. There was no blood in his stools and he made a full recovery, but the rash on his foot persisted and developed its serpiginous appearance by day 24. He was referred to ACU by his GP on day 27.

Blood tests at that time were unremarkable including normal white cell count (WCC; $8.4 \times 10^9/L$) and differential (eosinophil count: $0.2 \times 10^9/L$ (2%)), slightly elevated C reactive protein (CRP; 26.6 mg/dL) and normal renal and liver function. Stool samples were sent for microscopy, culture and sensitivity, including examination for ova, cysts and parasites (though his bowel habit had returned to normal). No enteric pathogens or parasites were identified. Blood cultures, urinalysis, malaria rapid diagnostic test, viral throat swab and a serology screen (HIV, syphilis, cytomegalovirus, Epstein-Barr virus) were all negative. Specialist serological tests were also sent for alphavirus, flavivirus, phlebovirus, rickettsia and leptospira at the Rare and Imported Pathogens Laboratory in Wiltshire, UK and were all negative. Given the distinctive rash appearance and lack of any invasive disease, he was diagnosed clinically with CLM and sent home with a single dose of 200 µg/kg ivermectin, which he took the following day.

While his left foot lesions improved over the next few days, he became systemically unwell. The night of this attendance in ACU (day 27), he developed dyspnoea, tachypnoea, a productive cough, fever,

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Figure 1 Rash on patient's left foot at initial presentation. Photographs of patient's left foot (A), showing the two discrete lesions (B,C) numbered 1 and 2 in (A). The serpiginous, pruriginous erythematous tracks are characteristic of cutaneous larva migrans caused by non-invasive zoonotic hookworm species but can less commonly be associated with cutaneous gnathostomiasis.

night sweats and chest pain. He had these symptoms for 4 days before visiting his GP, who prescribed a 3-day course of amoxicillin, and his symptoms improved. Around day 34, his right foot became erythematous, painful and swollen, and he developed further swollen, erythematous lesions behind his right knee and calf. He described lesions appearing and disappearing around his right leg over the course of a single day. Blood tests showed a rising eosinophilia, with eosinophil counts of $0.7 \times 10^9/L$ (8%) on day 32 up to $1.4 \times 10^9/L$ (12.3%) by day 35 (though overall WCC remained within normal range). A chest X-ray (figure 2) from day 32 showed bilateral widespread multifocal airspace changes with a small left-sided pleural effusion. He was seen in the Rapid Access Infectious Diseases clinic on day 38, by which time he was clinically improving. The differential included larva *currens* (*Strongyloides*), visceral larva migrans, toxocariasis and gnathostomiasis. He was treated with a further two doses of 200 µg/kg ivermectin taken 24 hours apart and prescribed a 7-day course of albendazole 400 mg two times per day (though the patient later reported he did not take this). Blood samples sent to the Department of Parasitology, Hospital for Tropical Diseases, London, were negative for serological evidence of *Toxocara* or *Strongyloides*. However, the laboratory in Mahidol University in Bangkok returned a positive result for the *Gnathostoma spinigerum* immunoblot, confirming the diagnosis of gnathostomiasis.

Figure 2 Chest radiographs on re-presentation with respiratory symptoms. Chest radiographs from day 32 after leaving UK (A) and 16 days later (B). (A) Bilateral widespread multifocal airspace changes with a small left-sided pleural effusion, presumably parapneumonic. (B) Almost complete resolution of pathological features from radiograph (A). Transient pulmonary infiltrates with rapid resolution on chest radiography is a distinguishing feature of Löfller's syndrome.

DIFFERENTIAL DIAGNOSIS

Differential diagnosis at time of re-presentation with respiratory symptoms:

- ▶ gnathostomiasis
- ▶ toxocariasis
- ▶ larva *currens* (*Strongyloides*)
- ▶ CLM with Löfller's syndrome
- ▶ visceral larva migrans.

TREATMENT

Initially, the patient received a single dose of 200 µg/kg ivermectin after his initial presentation with CLM-like lesions. He had a second treatment after re-presenting with Löfller's syndrome and migratory swellings, consisting of two 200 µg/kg ivermectin doses taken 24 hours apart. He was also prescribed 7 days of two times per day 400 mg albendazole but did not take it. After the relapsing cutaneous lesions were discovered he was given a 21-day course of two times per day 400 mg albendazole.

OUTCOME AND FOLLOW-UP

A repeat chest X-ray on day 48—16 days after the first chest X-ray demonstrating pulmonary infiltrates—had almost entirely returned to normal (figure 2). At a clinic review on day 74, all symptoms had fully resolved. His eosinophil count reduced over several weeks, at first still occupying a high proportion of total white cells ($1.1 \times 10^9/L$, 81.9%) but ultimately normalising.

However, 36 weeks after he was first seen in ACU, he reported recurrence of skin lesions, with swellings in his cheek and chin appearing every 2–3 weeks and lasting 3–4 days before spontaneously settling. There were no other symptoms and no eosinophilia. On examination, he had a ~1 cm lump on the side of his chin, which was warm with slight fluctuance. It was not itchy and there were no serpiginous tracks. He was given a 21-day course of two times per day 400 mg albendazole, and on follow-up 8 weeks later reported no recurrence of his symptoms.

In summary, this was a previously fit and well 32-year-old man who first presented systemically well with skin lesions typical of CLM following a holiday in South-East Asia, and was treated with a single dose of ivermectin. He then developed respiratory symptoms and became systemically unwell, with peripheral eosinophilia and multifocal airspace changes in both lungs. This episode resolved but he suffered recurrent migratory erythematous skin swellings and was seropositive for *G. spinigerum*.

DISCUSSION

This patient's initial presentation was classic for CLM: systemically well with the distinctive serpiginous rash (figure 1) following recent travel to South-East Asia, including time spent barefoot in areas with stray dogs and cats. CLM is a self-limiting dermatological condition resulting from the presence of zoonotic nematode larvae which are limited to the epidermis. The most common cause of CLM is dog and cat hookworm species (eg, *Ancylostoma braziliense* and *Ancylostoma caninum*) acquired in tropical regions^{4–7} and associated with walking barefoot.⁸ The larvae are unable to penetrate into deeper tissues, causing characteristically pruriginous, serpiginous 'creeping eruptions' in the skin, most commonly in the lower extremities, especially the feet.⁹ In contrast, cutaneous gnathostomiasis typically presents as intermittent migratory swellings which can be pruritic, painful and erythematous.¹⁰ We cannot exclude the possibility that our patient was simultaneously coinfecting with both zoonotic hookworm, causing his initial CLM lesions, and *Gnathostoma* larvae, causing the subsequent respiratory illness and migratory

swellings. However, creeping eruption skin lesions that mimic CLM have been described uncommonly in gnathostomiasis.¹⁰ In a study of 78 patients with creeping eruption seen in Pitié Salpêtrière Hospital in Paris, the vast majority were associated with hookworm infection, with 2/78 (3%) attributed to *Gnathostoma*.¹¹

Larva currens also features linear, serpiginous cutaneous lesions, caused by transdermal migration of the nematode *Strongyloides stercoralis*. A distinguishing feature is the speed of the lesions' migration. First reported by Arthur and Shelley in 1958, they vividly described how, 'It was possible actually to see advancement of the lesion within a matter of 10 minutes. After a period of hours the lesion would cease moving, only to resume movement at an unpredictable time the following day'.¹² They named it 'larva currens' or 'racing larva'. Our patient's *Strongyloides* serology was negative, and his lesions did not have the typical urticarial appearance associated with migrating *Strongyloides* larvae.

The patient then developed respiratory symptoms, peripheral eosinophilia and transient pulmonary infiltrates (with radiological resolution within 16 days of appearance on chest X-ray)—the classic triad of Löffler's syndrome.¹³ A distinguishing feature of the syndrome identified by Wilhelm Löffler in 1932 was its transience: 'Decisive is the rapid development of both, the pulmonary infiltrations and the blood eosinophilia', with X-ray findings usually appearing within 3 days and disappearing in 10–12 days.¹⁴ Löffler's syndrome is most commonly associated with invasive anthrophilic helminths such as *Ascaris*, which have pulmonary migration phases in their life cycles.^{15–16} Two mechanisms that have been proposed^{17–18} for the pathogenesis of Löffler's syndrome are: direct damage if the nematode larvae undergo pulmonary migration and a more systemic immunological reaction to helminth antigens, which typically trigger TH₂ responses associated with eosinophil activation.¹⁹ Cases of Löffler's syndrome complicating CLM have also been reported in the literature since Wright and Gold's 1946 case series.^{17–20–31} Our case raises the question of whether some of these were caused by *Gnathostoma* larvae rather than the typically non-invasive zoonotic hookworm species more commonly associated with CLM.

Worldwide, helminth infection is the most common cause of persistent eosinophilia.³² Other causes include allergy (asthma/atopy, drug hypersensitivity), haematological/neoplastic (leukaemia, lymphomas, mastocytosis), immunological (graft vs host disease, rare immunodeficiencies such as Hyper-IgE syndrome), endocrine (hypoadrenalism) and idiopathic.³² For helminth infections, contact with the host immune system is required to elicit the TH₂ immune response that drives eosinophilia.³³ This is generally only seen in invasive helminths with a tissue migratory phase (eg, trichinosis, ascariasis, filariasis, gnathostomiasis—see table 1 in Nutman³² for comprehensive list) and not in cases where the parasite resides outside of tissues, for example, intraluminal, such as adult tapeworm species.

The diagnosis confirmed serologically in this case was gnathostomiasis—a foodborne zoonosis endemic in tropical regions where people eat freshwater fish or shellfish, particularly in South-East Asia and Latin America.¹⁰ *Gnathostoma* are parasitic nematodes with a complex life cycle that can infect a wide range of hosts (first discovered within the stomach of a tiger in London Zoo). Larvae shed in the faeces of definitive hosts (such as cats and dogs, which eat freshwater fish) develop within copepods (microscopic crustaceans) before being consumed by the second intermediate hosts—fish, eels, amphibians, birds and reptiles. The larval worms migrate through, and encyst within,

the tissues of these hosts, awaiting their ingestion by definitive hosts to complete the life cycle. Humans are accidental hosts; it is thought the main mechanism of infection is ingestion of undercooked second intermediate hosts (ie, infected freshwater seafood and other animals such as frogs, snakes and chicken). Our patient had stayed in coastal tropical regions eating local seafood and chicken.

The many case reports of human gnathostomiasis reflect the clinicopathological diversity of this protean parasitic illness, which can manifest with several cutaneous forms,^{34–38} ocular infection^{39–45} and visceral disease affecting the CNS,^{46–52} spine,^{53–55} lungs,^{56–58} gastrointestinal tract⁵⁹ and other tissues (further references in¹⁰). The range of organs affected reflects the ability of the mobile third-stage nematode larvae to migrate through and into different anatomical regions.⁶⁰ Asymptomatic eosinophilia has also been reported.⁶¹

Gnathostomiasis is most commonly encountered in European and North American healthcare settings in returning travellers from South-East Asia⁶² and South and Central America,^{63–64} though imports from Africa⁶⁵ and an endemic case in Australia⁶⁶ have been reported. The major risk factor for gnathostomiasis is consumption of raw freshwater seafood in endemic regions.⁶⁷ The increasing demand for 'exotic' foods including raw seafood dishes in formerly non-endemic countries has also led to autochthonous infections in people without significant travel history^{67–69}; for example, there have been gnathostomiasis cases reported in a habitual sushi eater from San Francisco⁷⁰ and a New Yorker who handled fresh fish in Chinatown.⁷¹

The clinical history of migratory cutaneous lesions, recent travel to endemic countries and/or ingestion of raw or undercooked seafood and other meats and peripheral eosinophilia should raise concern for gnathostomiasis.^{10–62} Diagnostic tests include seropositivity against a 24 kDa *G. spinigerum* L3 antigen⁷² and histopathological analysis of cutaneous biopsies, though the latter is complicated by the migratory nature of the skin lesions³⁶ and has become less common as serological testing

Learning points

- ▶ Gnathostomiasis can cause cutaneous larva migrans-like cutaneous manifestations in addition to the more typical migratory swellings (our patient had both). Several helminth species can cause distinct but overlapping spectra of cutaneous manifestations; in some instances, as in this case, distinguishing between the causal organisms can be initially challenging.
- ▶ Gnathostomiasis should be considered as a diagnosis in all patients who present with the triad of peripheral eosinophilia, migratory cutaneous lesions and recent travel to endemic areas and/or ingestion of undercooked seafood and meats.
- ▶ Gnathostomiasis and other helminth infections are increasingly seen in non-endemic countries due to greater international travel and autochthonous infections from 'exotic' foods such as undercooked seafood.
- ▶ Visceral gnathostomiasis can include a Löffler's syndrome-type illness of respiratory symptoms, transient pulmonary infiltrates and peripheral eosinophilia.
- ▶ Achieving long-term cure in gnathostomiasis can be challenging with a tendency for relapse months after treatment. Published studies to date have small sample sizes and further research is needed to assess optimal therapy for relapses and treatment efficacy for achieving long-term cure.

became available.¹⁰ Among samples sent to the Hospital for Tropical Diseases in Bangkok from 2000 to 2005 for *Gnathostoma* serology, seropositivity was around twice as likely for both a history of cutaneous swellings and of eating raw meat.⁷³

Long-term follow-up studies have shown a high prevalence of treatment failure in imported gnathostomiasis, echoed in our case. In one case series, 8 out of 13 patients relapsed a median of 2 months post-treatment, sometimes relapsing multiple times.⁷⁴ There are limited data suggesting that 21 days of albendazole is more effective than single dose 200 µg/kg ivermectin for inducing cure in gnathostomiasis, though with only 31 trialists, this study was underpowered and the difference was not statistically significant.⁷⁵

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Contributors WLH and DA were both involved in the patient's clinical care. DA was senior clinician. WLH and DA wrote the manuscript.

Competing interests None declared.

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