

## First reported case of intraocular *Gnathostoma spinigerum* in Cambodia

### Premier cas de gnathostomose intra-oculaire à *Gnathostoma spinigerum*, diagnostiqué au Cambodge

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**Abstract** A live intraocular nematode was identified from a 37 year-old man presented with iritis, pain, redness, lacrimation, swelling, vision loss and intermittent blindness during many hours per day of the left eye. By using slit lamp examination, a worm was removed from iris in an ophthalmology outpatient department setting and sent to the Medical Microbiology Laboratory, Institut Pasteur du Cambodge. *Gnathostoma spinigerum* was identified, based on its typical morphology via microscopic examination. Based on our diagnosis, the patient was treated by oral albendazole and responded well to this therapy.

**Keywords** Gnathostomiasis · *Gnathostoma spinigerum* · Diagnosis · Treatment · Hospital · Province of Kampong Cham · Phnom Penh · Cambodia · South East Asia

**Résumé** La gnathostomose est une helminthose tropicale animale due à un nématode du genre *Gnathostoma*. Ce genre comprend plusieurs espèces, dont la plus répandue est *Gnathostoma spinigerum*. Le cycle parasitaire est complexe et comporte au moins 3 hôtes avec la possibilité d'hôtes paraténiques. Le premier hôte intermédiaire est un crustacé (cyclops), le deuxième un poisson d'eau douce ou un batracien, tous les vertébrés peuvent être hôte paraténique, en particulier le porc et les oiseaux. L'Homme se contamine en consommant la chair crue ou peu cuite des hôtes intermédiaires ou paraténiques. Le plus souvent il s'agit de poissons d'eau douce, de porcs, mais aussi d'autres hôtes intermédiaires (serpents, anguilles...) contenant la larve infestante au troisième stade (L3). La contamination par pénétration transcutanée des larves L3 est possible, le temps

moyen de pénétration étant de 30 minutes avec un minimum de 6 minutes. Chez l'Homme comme chez les hôtes paraténiques le cycle est bloqué, c'est une impasse parasitaire : la larve L3 « erre » dans les tissus, responsable d'un syndrome de *larva migrans* cutanée et/ou viscérale. Nous rapportons le premier cas documenté de gnathostomose intra-oculaire survenu au Cambodge. Il s'agit d'un homme de 37 ans présentant des épisodes de cécité de l'œil gauche, avec douleurs, iritis, larmolement et œdème. L'examen à la lampe à fente a permis de mettre en évidence une larve de nématode, dont l'aspect était très évocateur de *Gnathostoma spinigerum* : tête typique en forme d'ampoule avec cinq rangées de petits crochets et fines épines cuticulaires sur sa surface corporelle. La mise en évidence de la larve a permis de faire le diagnostic de certitude. Lorsque la larve n'est pas identifiée, le diagnostic repose le plus souvent sur un faisceau d'arguments épidémiologiques, cliniques et biologiques (hyperéosinophilie, sérodiagnostic positif). Il s'agit du premier cas intra-oculaire signalé au Cambodge. Cette pathologie ne doit pas être ignorée car la gnathostomose est une des causes de méningo-encéphalite à éosinophiles. Avant l'utilisation des antiparasitaires, l'atteinte neurologique était compliquée d'une létalité atteignant 25 % et de séquelles à long terme dans 30 % des cas.

**Mots clés** Gnathostomose · *Gnathostoma spinigerum* · Diagnostic · Traitement · Hôpital · Province de Kampong Cham · Phnom Penh · Cambodge · Asie du Sud-Est

## Introduction

Gnathostomiasis is a parasitic infection caused by ingestion of a third-stage (L3) nematode larva named *Gnathostoma spinigerum*, most commonly found in tropical and subtropical regions [6]. It has been reported widely in Japan and Southeast Asia in particular in Thailand and recently in India [5,10,11,17]. Infection may be acquired by drinking

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contaminated water or eating partially cooked fish or meat [15]. Gnathostomiasis causes lesions in various organs but the most common manifestation of infection involves the skin and subcutaneous tissues. Parasitic ocular disease caused by live *Gnathostoma spinigerum* is rarely diagnosed in humans, and may occur only after a certain time lapse after initial infection [6]. Gnathostomiasis is potentially serious with severe consequences; it is a cause of eosinophilic meningoencephalitis. Before the use of antiparasitic treatment, neurological involvement was complicated by up to 25% lethality and a large number of survivors were left with disabling neurological sequelae (in 30% of cases). To our knowledge, we report the first case of intraocular *Gnathostoma spinigerum* diagnosed in Cambodia.

## Case presentation

A 37 year-old man from southeastern Cambodia province of Kampong Cham self-referred to the ophthalmology outpatient department of the provincial hospital with a complaint of iritis, ocular pain, redness, lacrimation and swelling for one month, resulting in vision loss and intermittent blindness of the left eye during several hours per day for one week. There were no symptoms in the right eye.

The patient is a farmer, non-vegetarian and lives in poor home hygiene conditions in a farm near cattle, dogs and cats. When asked, he did not specifically report eating raw or undercooked meat or fish and drinking natural water. He reported no travel outside Kampong Cham province during the previous year, especially no travel to Thailand. His symptoms had appeared one month earlier. Clinical examination found no lymphadenopathy, edema or fever. In contrast, he complained of a severe headache. He had sought treatment because of intermittent blindness. Before referring to the ophthalmology department of a hospital in Phnom Penh, he consulted a general practitioner in his home province and received unidentified anti-inflammatory and antibiotic treatment. Slit lamp examination of the left eye at the ophthalmology department identified a live worm piercing the iris. The worm was removed by paracentesis of the anterior chamber, placed in a normal saline and forwarded to the Medical Microbiology laboratory, Institut Pasteur du Cambodge, Phnom Penh.

## Laboratory diagnosis

The worm underwent direct morphological examination in our laboratory under low and high power microscope magnification. It was reddish white in color and was measured as being 12.5 mm long and 1.2 mm wide (Fig. 1). It had a



**Fig. 1** *Gnathostoma spinigerum*. Photo credit: Alexandra Kerléguer, MD (IPC)

typical head bulb with five rows of hooklets and fine cuticular spines on its body surface (Fig. 2).

A blood cell count was performed which was normal except for a slight increase in the eosinophil count (7%). Eosinophilia is seen especially in association with helminth infections, and during the tissue-invasive stages of development.

Based on our finding, the worm was identified as a larva stage 3 of *Gnathostoma spinigerum*.

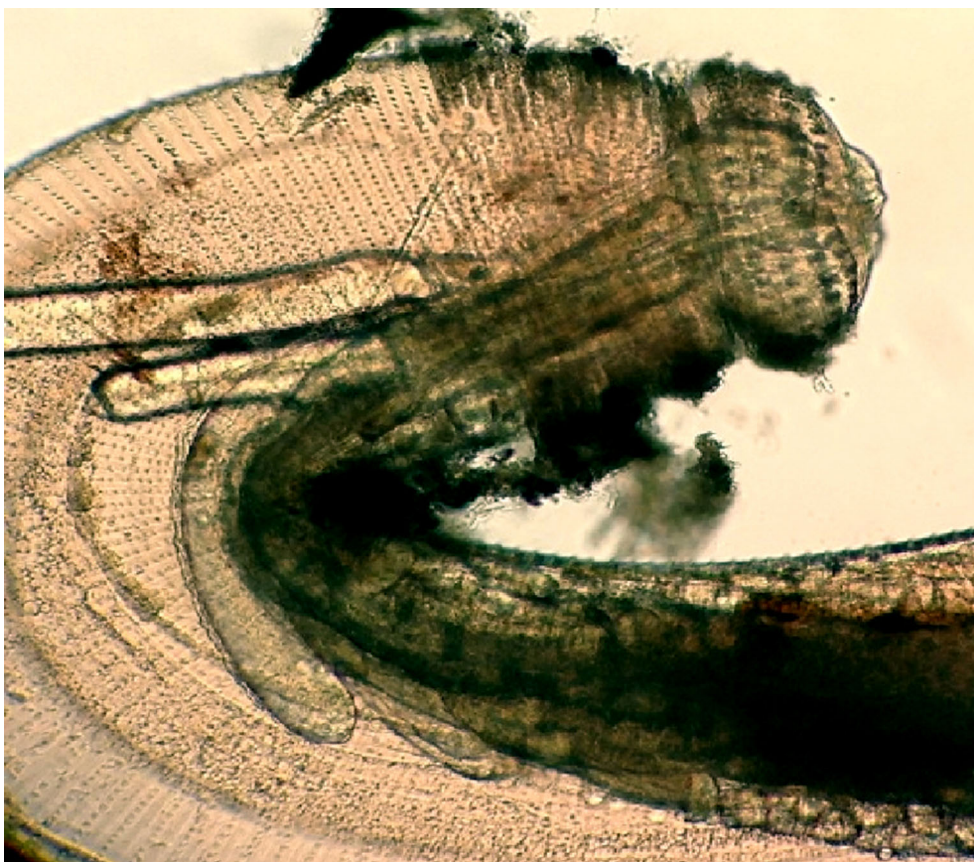
Despite no obvious epidemiological context, the typical appearance of the worm allows the diagnosis.

Clinically, according to the endemic areas, the main differential diagnoses include angiostrongyliasis, trichinellosis, and larva migrans (Loa-Loa, *Dirofilaria*, and *Onchocerca*).

The patient was treated with oral albendazole 400 mg daily for 3 weeks and responded well to the treatment. At the end of treatment, all symptoms had disappeared, including blindness episodes.

## Discussion

Human cases of live intraocular *Gnathostoma* are rare occurrence [14]. Gnathostomiasis is due to a human infestation by



**Fig. 2** Cephalic part of the worm / *Tête du nématode*. Photo credit : Alexandra Kerléguer, MD (IPC)

larvae of a nematode worm, *Gnathostoma sp.* usual parasite of dogs and cats. Its parasitic cycle is complex and involves two intermediate hosts: a freshwater crustacean and fish. Humans become infected with third-stage *Gnathostoma* larvae; worm parasites enter an organism through ingestion of raw or undercooked hosts, or rarely, through direct larval penetration of the skin [6,13]. L3 larvae of *Gnathostoma* penetrate the human gut and migrate through the peritoneal cavity to the liver. From there, they may migrate to subcutaneous tissues or various organs, as the central nervous system or the eye, without fully developing to adults worms [1,2,7]. The eye is the only organ in which the larva may be visualized [1,2,12]. Gnathostomiasis is mostly reported in Southeast Asia [5] but ocular involvement has never been reported before in Cambodia. Since the beginning of the eighties, there is an increasing number of cases of gnathostomiasis described in Western countries in travelers returning from endemic countries [4]. The most heavily endemic countries with human gnathostomiasis is Thailand, due to the eating habits [4,12].

During the invasion phase, clinical signs are not specific (malaise, fever, urticaria, nausea, anorexia, epigastric pain), reflecting the migration of the worm, but eosinophilia usually develops in association with larval penetration. The

larval worm then migrates to the skin through the subcutaneous tissue causing the typical migratory swellings. As the chronic stage begins and the larva enters the subcutaneous tissues, the eosinophilia usually subsides [3].

The key to the diagnosis of gnathostomiasis is based on anamnesis and clinical findings.

Laboratory testing is based on microscopic examination of larvae extracted from the lesions, serological tests (IgG ELISA, Immunoblot) or L3 antigen detection [9,16,18].

Treatment of gnathostomiasis is based on location of infection. For cutaneous disease, a 21-day treatment course of albendazole was found to be effective as a 21-day treatment of ivermectine [8,14], but albendazole is easy to find in Cambodia and less expensive. For parasitic ocular infection, the only treatment is the surgical removal.

This is the first case reported in Cambodia to the best of our knowledge. The incidence of this disease may be higher in a country like Cambodia because of conditions of poor hygiene and underdiagnosis by clinicians. The importance of gnathostomiasis should be recognized because of its potential severity.

This case underlines the need to inform travelers and migrants to endemic regions of the risks associated with



eating raw or undercooked fish and should be regarded as another emerging imported disease.

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Conflict of interest : No conflict of interest to declare

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