**Case Report** 

# Eyelid Gnathostomiasis Presenting as an Orbital Mass: A Case Report of Rare Presentation

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# Abstract

A 25-year-old female presented with a three-week history of mass in the left lower orbit. On examination, a well-circumscribed rubbery round mass protruding from the left lower eyelid was noted. An anterior orbitotomy was performed via transconjunctival approach. Ultimately, the diagnosis of gnathostomiasis was done pathologically.

Keywords: Gnathostomiasis, Orbital mass, Eyelid

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#### Introduction

The orbital mass has a wide variety of etiology, from a benign condition to malignancy in 6 main pathologic processes: inflammatory, infectious, hemorrhagic, neoplastic, metastatic, and developmental.<sup>1</sup> It requires complete history, physical examination, and further imaging for diagnosis. However, the diagnosis can be challenging and require tissue biopsy for a definite diagnosis. Gnathostomiasis is a food-borne parasitic disease. The endemic area is in Southeast Asia and Latin America, but the number has increased in Europe.<sup>2</sup> The parasite can migrate through human tissues, most commonly in skin and subcutaneous tissue.<sup>3</sup> However, it can penetrate the ocular tissue and may cause blindness.<sup>4</sup> We present a case of a young adult who presented with orbital mass visible at lower eyelid, diarrhea, and urticaria.

## **Case report**

A 25-year-old female doctor, who lived in suburban Bangkok, Thailand, presented at the Ophthalmology Department of Thammasat University Hospital with the chief complaint of a left lower eyelid mass for three weeks. She was previously prescribed Augmentin (1g) 1 tablet twice a day for seven days with minimal improvement.

Her best corrected visual acuity was 20/20. The left lower eyelid mass was approximately 2.5 cm in diameter. It was firm, round, non-pulsatile, and movable. The patient had no proptosis and no sign of periorbital change. The remainder of the exam was unremarkable. A complete blood count (CBC) had the following results: WBC 5,400 cells, neutrophil 61.4%, lymphocyte 24.0%, monocyte 9.4%, eosinophils 2.5%, basophil 0.7%, hematocrit 37.2%, hemoglobin 12.5g/dL, and platelet 237,000/µL. The anti-HIV was non-reactive. She also had preauricular lymphadenopathy with the reactive lymphoid population on the aspirate smear cytology. The CT scan shown below was performed (Figure 3A - 3D). An anterior orbitotomy with exploration and removal of the mass was performed. Pathological analysis of the mass revealed nematode larva surrounded by a foreign body granuloma. The worm was 0.24 mm in diameter and at least 5 mm in length showing cuticular spines, large cavernous lateral chords, and pigmented granules in the intestinal cells, suggestive of Gnathostoma. With a positive immunoblot for Gnathostoma, the diagnosis of gnathostomiasis was made. (Figure 4). The patient received ivermectin (6 mg) two tablets per day for two consecutive days. There was no recurrence at three weeks and two months follow-up visit.



Figure 1 A rubbery grey-white tissue measuring  $1 \times 1 \times 1$  cm<sup>3</sup>.

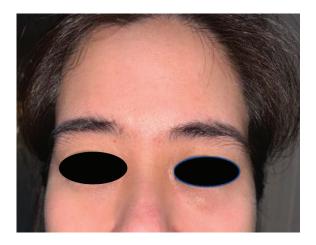


Figure 2 The left lower eyelid mass.

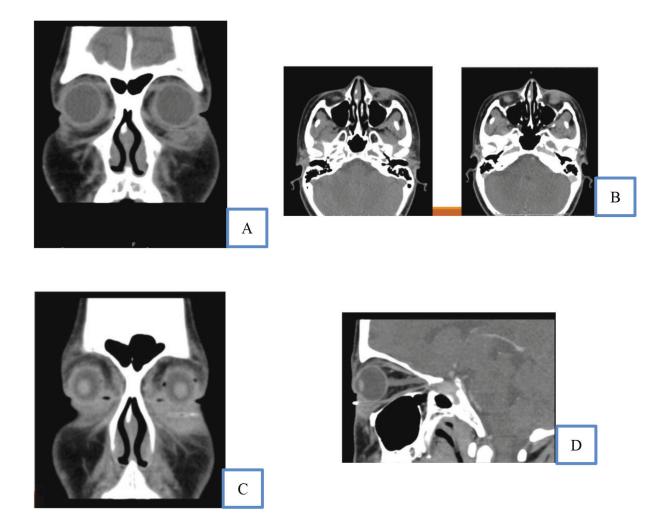


Figure 3 (A) CT orbit axial view shows a 3.1×1.4×1.9 cm, an ill-defined lesion with soft tissue density at the left lower eyelid, involving subcutaneous fat, facial muscles, and premolar fat, including anteroinferior extraconal fat of left orbit. (B) axial view without and with contrast, respectively. (C) coronal view with contrast. (D) left sagittal view with contrast.

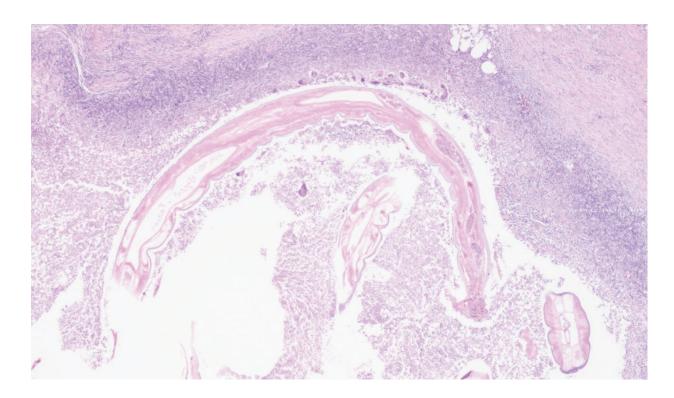


Figure 4 The mass contains few degenerating nematode larvae surrounded by foreign body granuloma  $(H\&E \times 20)$ .

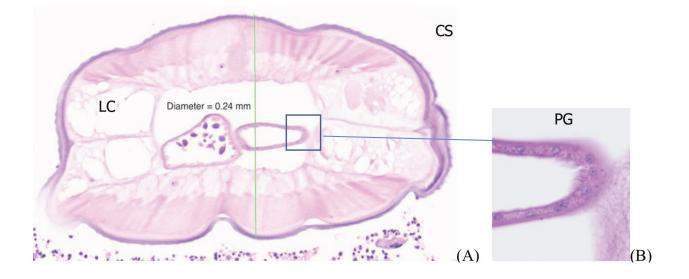


Figure 5 (A) The worm measures 0.24 mm in diameter and at least 5 mm long shows cuticular spines (CS) and large cavernous lateral chords (LC) (H&E × 100). (B) The intestinal cells have brown pigment granules (PG) (H&E × 400).

#### Discussion

Human gnathostomiasis is most commonly diagnosed in Southeast Asia. It is caused by parasitic worms in the genus Gnathostoma. Humans become infected by consuming undercooked fish, eels, birds, and reptiles. Although most infections are localized, the parasite can intermittently migrate and cause swelling of cutaneous tissues.<sup>5</sup> The most common manifestations of ophthalmic gnathostomiasis are anterior uveitis due to the localization in the anterior segment. Other ocular manifestations may include secondary glaucoma, retinitis, subretinal hemorrhage. The larvae may also cause orbital inflammation by traveling along the optic nerve as they enter the eye. Morphologic diagnosis requires identification of larvae in the skin biopsy, although rarely performed. In histologic sections, Gnathostoma spp. often can be differentiated from one another on the basis of the morphologic features of their intestinal epithelial cells. The serologic test using the 24 kDa of a specific reactive band in western blotting is particular. There is a case report of ocular gnathostomiasis in a sixteen-year-old Thai girl with an acute unilateral visual loss, proptosis, and ophthalmoparesis. Her serum was positive for Gnathostoma spinigerum using an immunoblotting technique.<sup>4</sup> In this case, the diagnosis was made from the anterior orbitotomy with a positive immunoblot for Gnathostoma. The treatment of choice is oral albendazole 400 mg bid for 21 days or a single dose of ivermectin 0.2 mg/kg that may be repeated after seven days.<sup>6</sup> The patient underwent an excisional biopsy and was treated with ivermectin (6 mg) two tablets for two consecutive days. Drug re-administration may be considered if recurrence occurs. The reinfection of cutaneous manifestation usually does not occur until four weeks after the initial infection. In this case, there was no new

mass, no recurrence, and no flare at the follow-up visit at three weeks and two months. The patient was determined to be cured of gnathostomiasis.

We encountered a rare presentation of gnathostomiasis, which is an orbital mass. Therefore, once the patient comes with systemic presentation including urticarial rash and diarrhea, along with an orbital mass, gnathostomiasis should be considered as one of the differential diagnoses.

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