Intravitreal Angiostrongyliasis : Report of 2 Cases†

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Abstract

Two male patients presented with unilateral blurred vision. A small motile worm was found in the vitreous cavity in both cases. In one case, another dead, disintegrated worm was noted in the inferior portion of the vitreous cavity. On each eye, vitreous surgery was performed and the worm was removed by an aspirator. Both worms were identified as *Angiostrongylus cantonensis*. Since both patients had no signs of meningitis, lumbar puncture was not done. Long-term follow-up confirmed the benefit and safety of the vitreous surgery. Fluorescein angiography revealed severe pigmentary alteration, probably from inflammation of the choroid and retina due to subretinal migration of the worm prior to access into the vitreous cavity.

Key word : Angiostrongylus, Vitrectomy

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Angiostrongylus cantonensis, a rat lungworm, is well known as an etiologic agent of eosinophilic meningitis commonly found in many Pacific islands and southeast Asia[1,2]. Adult worms reside in the pulmonary arterioles of rats and certain other rodents, where ova hatch to the first-stage larva. The larvae then migrate to the bronchi and esophagus, and are passed in the feces. Snails, slugs, and crustaceans are intermediate hosts. When rats eat these organisms, third-stage infective larvae enter their circulation and

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reach the central nervous system, where they develop into the fourth and fifth larval stages. They then enter the venous circulation again and return to the heart, and finally mature to adult worms in the rat's pulmonary arteries. Humans are facultative hosts and become infected by ingesting raw intermediate hosts or by consuming contaminated water or vegetables[3,4]. In humans, the larvae migrate through the circulation to the brain where they grow to fourth and fifth stage larvae. Rarely, they may travel through the optic nerve and penetrate into the eye. Although it is estimated that thousand cases of eosinophilic meningitis occur annually, ocular involvement is rare[4].

There have been 12 reports of ocular angiostrongyliasis in eleven English[5-15] and Thai[16] literature. The location of worms was in the anterior chamber (5 cases)[5,7,10,11,16], subretinal space (2 cases)[9,14], and vitreous cavity (4 cases)[6-8,15]. In another case, two worms presented in the same eye; one in the anterior chamber and the other in the vitreous cavity[12]. The patients may or may not have accompanying clinical evidence of central nervous system involvement[8,14]. Of 5 intravitreal cases, two worms were removed and histologically confirmed [6,8]; two of the remaining three were speculated by serum antibody tests[7,12,15].

The natural history of intraocular Angiostrongylus is not known. If the location of the worm can be easily accessed such as in the anterior chamber, it should be removed as performed in most reported cases[7,10,11,16]. There has been a report that a small motile worm just beneath the posterior vitreous face which ended up with good anatomical visual outcome without removal[15]. However, there was a report that a living worm in the subretinal space could lead to panuveitis, disc edema and retinal detachment requiring vitrectomy procedure[14]. The authors report 2 cases of intravitreal angiostrongyliasis-clinical manifestation, management, and long-term anatomical and visual outcomes.

Case 1

A 28-year-old man was admitted to the Maharaj Nakorn Chiang Mai Hospital on November 12, 1999. He complained of progressively decreased vision in his right eye for 2 weeks. A few days before the onset of blurred vision, he noticed a small, painless subcutaneous lump at the temporal aspect of the ipsilateral eyebrow. The mass spontaneously disappeared shortly before the onset of visual symptom. He experienced no headache, nausea, fever or other symptoms indicating meningitis. Eating snails was not his habit but raw meat and vegetables were often served at the table.

Examination of the left eye revealed no abnormality and visual acuity of 20/20. The right eye had a visual acuity of counting fingers at 1 foot; there were 1+ cells in the anterior chamber and 2+ cells in the vitreous cavity. A mild degree of relative afferrent pupillary defect was noted. There was severe, generalized pigmentary changes of the retina and choroid. A motile worm slightly longer than 10 mm was found in the inferior part of the vitreous cavity. Blood examination revealed a white blood count of 13,200/mm^3 with 5 per cent eosinophils. Since he had nothing suggesting meningitis, lumbar puncture was not done. Three days later, he underwent a pars plana vitrectomy procedure. Fig. 1 shows the intra-operative view. The worm was removed by a manual active aspirator. An assistant exerted a negative pressure on a 10-ml disposable syringe connected to an extension tube and a blunt-tip cannula to catch the worm. The parasitological report identified an immature adult *Angiostrongylus cantonensis*. Post-operatively, the intraocular inflammation rapidly decreased. At day 10, his visual acuity returned to 20/200. Twenty-eight months after surgery, his visual acuity was stable at 20/200 and the pigmentary retinopathy persisted.

Case 2

A 44-year-old man presented with blurred vision in his left eye. He denied eating snails but often had raw meat and vegetables. He had no symptoms and signs of meningeal involvement. Eye examination showed visual acuity of hand motion and 1+ cells in the anterior chamber as well as in the vitreous cavity. The intraocular pressure was normal. The fundus showed severe pigmentary changes and an active motile worm was seen in the vitreous cavity and superonasal to the optic disc (Fig. 2). Another dead, partially disintegrated worm was also noted in the inferior part of the vitreous cavity. The macula was mildly edematous. Mid phase of the fluorescein angiogram showed multifocal areas of hypofluorescence and hyperfluorescence throughout the fundus corresponding to pigment alteration seen on the fundus examination (Fig. 3A). There was slight leakage along the inferotemporal retinal vein and no cystoid macular edema in the late phase of the angiogram (Fig. 3B). The right eye was unremarkable. He underwent a pars plana vitrectomy using the technique described in case 1. The living worm was removed and noted to be

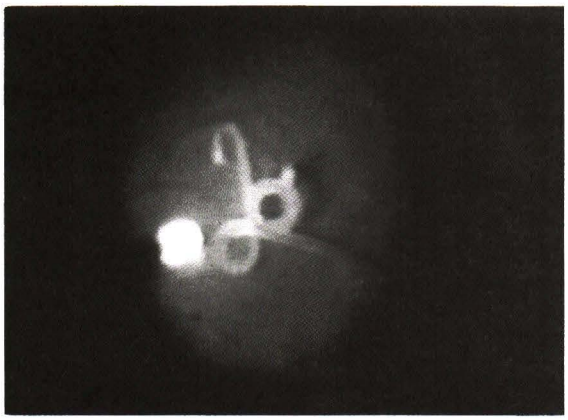

Fig. 1. An intra-operative view, a motile worm in the vitreous.

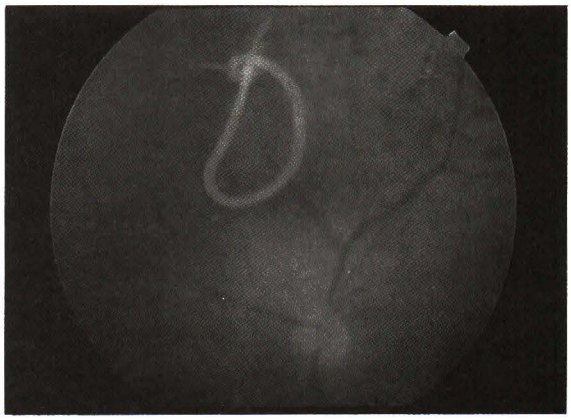

Fig. 2. The fundus shows severe pigmentary changes and an active motile worm can be seen superonasal to the disc.

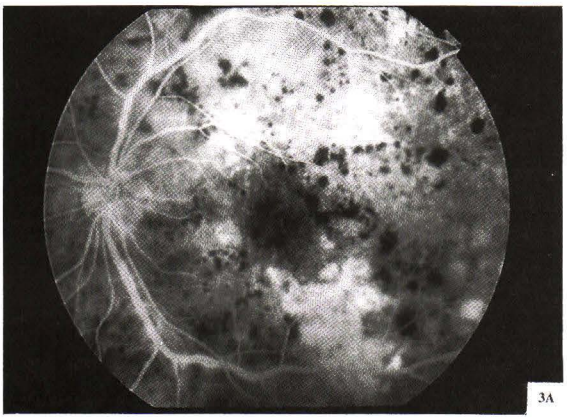

Fig. 3A. At 55 seconds after dye injection, fluorescein angiogram shows multifocal areas of hyperfluorescence and hypofluorescence, resulting from severe pigmentary alteration of the retina and choroid.

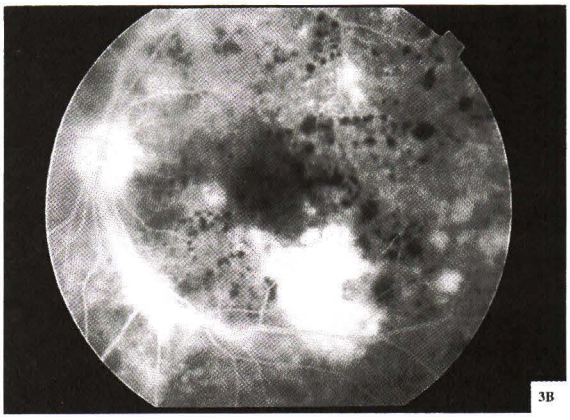

Fig. 3B. At 8 minutes after dye injection, slight leakage along the inferotemporal retinal vein was noted.

about 12 mm in length. The other dead worm was removed by the vitrectomy cutter. The living worm was sent to the Department of Parasitology for identification and reported to be an immature male *Angiostrongylus cantonensis*. The peripheral white blood count was 7,200 cells/mm^3 with a differential of 5 per cent eosinophils, 60 per cent neutrophils, and 32 per cent lymphocytes. Lumbar puncture was not performed. By the fourth week after surgery, visual acuity improved to 3/200; there were no inflammatory cells in the anterior chamber and vitreous cavity, and the macular edema resolved but pigment dispersion of the choroid and retina persisted. At his final visit, 13 months later, the visual acuity was 6/200.

DISCUSSION

As the present report cases had not eaten snails, fresh vegetables contaminated by larvae dis-

persed from infected snails is a possible source of infection. There have been reports that intraocular Angiostrongylus can lead to visual impairment secondary from panuveitis[14], macular edema[5], disc edema[14], severe pigmentary chorioretinopathy[4], and retinal detachment[8,14]. This present report showed low-grade panuveitis, severe pigmentary alteration of the choroid and retina. Fluorescein angiographic study revealed mild retinal vascular leakage, and multifocal hypo- and hyperfluorescence secondary to retinochoroidal inflammation. The pigmentary alteration probably resulted from inflammatory response while the worm migrated in the subretinal space prior to migration into the vitreous cavity[8,9].

The authors found that removal of the worm from the vitreous cavity facilitated amelioration of intraocular inflammation and visual recovery as seen in previous reports[6]. As a result, it may prevent more severe retinal, macular, and optic nerve damage from ongoing inflammation. Additionally the worm could be proved histologically[6,8]. The authors believe that severe pigmentary changes of the retina and choroid contributed to poor final visual outcome in the presented cases. Because the disease pathology relates to inflammation, early presentation, diagnosis and treatment before development of permanent damage from the severe inflammatory response of the choroid and retina may achieve better visual outcome. The presented cases did not shows overt clinical manifestation to central nervous system involvement, and a lumbar puncture was not performed.

SUMMARY

Intravitreal angiostrongyliasis can be seen with no accompanying clinical evidence of central nervous system involvement. Removal of the worm from the vitreous cavity is recommended since intraocular inflammation rapidly decreased after surgery. Early presentation and treatment are likely to have optimal visual outcome. However, avoiding servings of raw food will prevent this occurrence.

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พยาธิแองจิโอสตรองจิลัสในน้ำวุ้นตา : รายงานผู้ป่วย 2 ราย†

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ผู้ป่วยชาย 2 รายมาพบแพทย์ด้วยอาการตามัวข้างเดียว ตรวจร่างกายพบว่ามีพยาธิที่ยังมีชีวิตหนึ่งตัวอยู่ในน้ำวุ้นตาทั้งสองราย และหนึ่งในสองรายนี้ยังพบมีพยาธิที่ตายแล้วอยู่ในทางด้านล่างของช่องน้ำวุ้นตาอีกหนึ่งตัวด้วย ทั้งสองรายได้รับการผ่าตัดน้ำวุ้นตาเพื่อเอาพยาธิออก จากการตรวจทางพยาธิวิทยาพบว่าเป็น *Angiostrongylus cantonensis* เนื่องจากทั้งสองรายนี้ไม่มีอาการของเยื่อหุ้มสมองอักเสบจึงไม่ได้รับการเจาะตรวจน้ำไขสันหลัง จากการติดตามผลระยะยาวพบว่าการผ่าตัดน้ำวุ้นตามีความปลอดภัยและมีประโยชน์ช่วยให้การมองเห็นของผู้ป่วยดีขึ้นทั้งสองราย ในการตรวจโดยการฉีดสีฟลูออเรสซีนและถ่ายรูปจอประสาทตาพบว่าจอประสาทตามี pigment ผิดปกติอย่างรุนแรง น่าจะเกิดจากการอักเสบของฆอรอยด์และจอประสาทตาในขณะที่พยาธิไชอยู่ใต้จอประสาทตาก่อนที่จะเคลื่อนเข้ามาอยู่ในช่องน้ำวุ้นตา

คำสำคัญ : พยาธิแองจิโอสตรองจิลัส, การผ่าตัดน้ำวุ้นตา

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