Intraocular paragonimiasis

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SUMMARY A case of intraocular paragonimiasis is reported in a 13-year-old Chinese boy. The disease manifested as repeated attacks of acute intraocular pain associated with panuveitis. A combination of inflammatory reaction and ocular findings mimicking both perforating and contusion injuries caused by the migration of the fluke within the eye characterises the infection. The living fluke was successfully extracted from the anterior chamber and identified as *Paragonimus westermani*.

Paragonimiasis is a chronic lung infection endemic to the Far East which results from the ingestion of raw crustaceans contaminated by the trematode *paragonimus*. Extrapulmonary foci including the abdomen, brain, muscle tissue,¹ and temporal bone² have been reported. Ophthalmic manifestations of the disease include invasion of the subconjunctival space, eyelid, or orbital tissue by the worm as well as symptoms resulting from cerebral infection.³⁻¹⁰ Intraocular involvement was first reported by Jiang and coworkers, who described *paragonimus* worms in the anterior chamber in 3 of their 7 cases.¹¹ We believe the present case to be the first report of intraocular paragonimiasis in the Western literature.

Case report

A 13-year-old boy from the Chekiang province of the People's Republic of China presented with a 3-week history of decreased visual acuity OD accompanied by repeated attacks of severe ocular pain. He had been punched in the right temporal region one month before presentation. The blow caused no immediate eye symptoms, but the patient did experience some pain and hearing loss in the right ear. The following day the right lid was swollen, but no other ocular symptoms were present. The swelling persisted for 2 weeks and was followed by localised conjunctival congestion in the temporal aspect OD. Several days

later the patient experienced acute pain OD accompanied by metamorphopsia and decreased vision. These symptoms intensified with time. A diagnosis of glaucoma and intraocular haemorrhage was made, and the child was referred to the Eye, Ear, Nose and Throat Hospital of the Shanghai First Medical College.

On admission on 26 July 1979 visual acuity in the right eye was hand movements. Conjunctival congestion and corneal oedema were noted. A bloody exudate filled the anterior chamber and obscured the fundus. The pupil was dilated. Intraocular pressure was 43.3 mmHg by Schiøtz tonometry. Vision in the left eye was 6/7.2 and examination was unremarkable. A tentative diagnosis of haemorrhagic glaucoma was made, and the patient was started on a regimen of acetazolamide and topical as well as systemic steroids.

One day after admission the patient complained of acute ocular pain. The anterior chamber of the right eye was filled with a yellow fibrotic exudate, and vision in that eye had decreased to light perception with poor projection. Intraocular pressure was 33 mmHg. A diagnosis of acute iridocyclitis with secondary glaucoma was considered at this time.

On 31 July, five days after admission, the corneal oedema had resolved, intraocular pressure was within normal limits, and the anterior chamber exudate was largely absorbed, revealing iris atrophy and an irregular pupil. The sphincter muscle was lacerated at 5 o'clock. The anterior chamber appeared to be

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relatively deep and the lens tilted backwards in the superonasal quadrant. Gonioscopic examination revealed that the angle had widened. A large amount of pigment was present on the inferior part of the trabecular meshwork. An iris hole was noted near the root at 2 o'clock. Vitreous haemorrhage was present, yet through less dense areas a retinal detachment in the inferotemporal quadrant was noted. Retinal haemorrhages and exudates were scattered over a large part of the observed retina.

X-ray films failed to demonstrate a metallic intraocular foreign body, and A-scan ultrasonography confirmed the presence of the retinal detachment without underlying solid mass. Suspicion of a traumatic aetiology was aroused.

The following day the patient was awakened by severe pain OD. An examination showed exacerbation of the intraocular inflammation. Intraocular pressure was 13.1 mmHg. Several similar attacks followed, yet no apparent causes of predisposing factors could be identified.

On 6 August severe pain returned OD with conspicuous inflammatory reaction. A yellowish exudate streaked with blood appeared in the anterior chamber. The blood could be traced to the superonasal iris, where an iris hole was noted. At 5 o'clock near the angle a white mass measuring 2×3 mm was observed. Under slit-lamp examination this was found to be a motile, ovoid worm. Within seconds the worm was observed to burrow into the iris at 4 o'clock and it disappeared into the posterior chamber. Initially an iris bulge could be seen; when this subsided an iris hole was present marking the

point of exit of the worm (Fig. 1). This sequence of emergence and disappearance through the iris was repeated several times over the next few days. The patient experienced acute pain OD without the reappearance of the worm, and although no analgesic

Fig. 1 Patient's right eye, showing iris hole burrowed by the worm at 4 o'clock (arrow) and posterior synechia at 12 o'clock with fibrous membrane on the anterior lens surface.

Fig. 2 Fixed *Paragonimus westermani* after extraction from the anterior chamber. Fixed dimensions were 1.5×5 mm (OS=oral sucker).

relieved the pain it would subside spontaneously, even in the absence of medication.

On 11 August the fluke reappeared in the anterior chamber in the 5 o'clock position. A limbal incision was made promptly and the worm successfully extracted. It was alive and active following extraction from the anterior chamber. The Department of Parasitology at the Shanghai First Medical College identified the worm as *Paragonimus westermani* (immature) (Fig. 2).

The patient had an uneventful postoperative course. There was no further ocular pain; and the anterior chamber reaction subsided. The iris was highly atrophic and showed many small holes in the 2, 4, 5, and 7 o'clock positions. The lens was subluxated with pigmentation on its anterior surface. The old vitreous haemorrhage became organised. The retina was totally detached and showed haemorrhages and proliferative changes. No retinal breaks were found. Intraocular pressure was low.

Following identification of the parasite the patient revealed that he frequently ate raw crab and had last done so 3 months previously. Paragonimiasis was known to be present in his village.

No ova could be identified in sputum or faeces from this patient. Chest x-ray showed no changes indicative of pulmonary paragonimiasis. The white cell count was $17.4 \times 10^9/l$, and no eosinophils were found on admission.

Discussion

In the life cycle of paragonimus, ova are released from ruptured cysts in the human host's lungs and leave the body in the sputum or faeces. Given an aquatic environment, the ova hatch into miracidia which parasitise snails. Within the snail the parasites develop into cercariae and emerge approximately 13 weeks after infection. The crab or crayfish becomes host to the cercariae, which encyst as metacercariae. Following ingestion of raw affected crustacea by man, the encysted metacercariae pass through the duodenal wall into the abdominal cavity. The majority of worms then burrow through the diaphragm, enter the pleural cavity, and finally reach the lung.

The clinical features of intraocular paragonimiasis, as shown by our case as well as the 3 previously reported cases,¹¹ are summarised in Table 1. Repeated attacks of severe intraocular pain and exudative uveitis with profound visual loss are characteristic. The pain lasts approximately 30 minutes in each attack, is not alleviated by analgesics, and subsides spontaneously. This course differentiates these attacks from those of any type of acute glaucoma or iridocyclitis, though uveitis and

secondary glaucoma are often found in this disease. Coincident inflammatory reactions rule out migraine. On several occasions the pain was associated with the observed emergence of the worm in the anterior chamber. The patient experienced no further intraocular pain after extraction of the worm. These findings suggest that the pain is the result of the migration of the fluke through the nerve-rich uveal tissue.

Panuveitis caused by intraocular paragonimiasis typically appears as an exudative form associated at times with blood. However, the best diagnostic sign which we observed is the presence of multiple intraocular injuries in addition to the severe inflammatory reaction. In our case traumatic findings included laceration of the sphincter muscle, iris holes, deep anterior chamber with subluxation of the lens, and vitreous and retinal haemorrhage. These findings appeared at first to be contradictory. They cannot be explained as endophthalmitis caused by penetrating injury or by blunt trauma associated with severe intraocular reactions. A definitive diagnosis could not be made until the worm emerged into the anterior chamber. In retrospect these bizarre findings can be understood as being the results of the migration of the living fluke causing mechanical trauma to the intraocular tissues. It is not known whether additional biochemical injury from substance(s) produced by the worm exacerbated the damage to the eye.

Secondary glaucoma is another major manifestation of this disease. It has been recorded in all reported cases. Both inflammatory reaction and traumatic injury of the anterior chamber angle tissue may obstruct aqueous outflow and raise the intraocular pressure. Early surgical extraction of the worm

Table 1 Clinical features of intraocular paragonimiasis

Case	1*	2*	3*	4†
Sex	F	F	M	M
Age	7	11	6	13
Laterality	OD	OD	OS	OS
Ocular pain	+	+	+	+
Visual acuity	Unknown	LP	NLP	HM
Exudation or hypopyon	+	+	+	+
Retinal haemorrhage	+	Unknown	Unknown	+
Vitreous haemorrhage	—	Unknown	Unknown	+
Intraocular pressure	T+2	49 mmHg	50 mmHg	43 mmHg
Surgical extraction of immature fluke	Yes	Yes	No	Yes
Intradermal test	+	+	+	Unknown

*Cases 1, 2, 3 from Jiang *et al.*¹¹ †Case 4 from present paper. LP=light perception. NLP=no light perception. HM=hand movements. All tensions measured by Schiøtz tonometry.

is mandatory both to preclude permanent damage and to reduce intraocular pressure.¹¹

Several possible routes of intraocular infection may be considered. Cerebral paragonimiasis has been frequently reported among cases of pulmonary infection and has a reported incidence of 0.8 to 26.6%.^{7,9,12} It has been suggested that larvae or adult worms gain entry to the cranial cavity via the perivascular tissue of the jugular vein.^{6,13} According to Oh,⁸ basal arachnoiditis is a common finding in cerebral paragonimiasis and may result in optic atrophy. One might speculate, then, that the fluke migrates in a retrograde fashion along the optic pathway and enters the eye through the optic nerve. The absence of cerebral symptoms in our case makes that route unlikely.

Animal experiments conducted by Mo¹⁴ showed penetration of the globe by paragonimus metacercariae placed in the conjunctival sac. Two weeks after infection immature flukes were found in the choroid, and the affected eye also showed limbal and scleral infiltration, uveitis, and uveal haemorrhage. These data suggest another possible route of infection in this case may be migration through the soft tissues of the neck, ultimately to the right upper lid. The swelling of that lid may be attributed to this invasion and support this theory. The role of the prior punch to the head is unclear, but it may have prompted the migration. From the lid the fluke may have invaded the subconjunctival space, where local congestion was noted. Finally, the scleral or limbal tissue may have been penetrated, and thus the worm became an intraocular resident. The active motility we observed both in vivo and in vitro makes this hypothesis plausible. Embolic dissemination in the egg or metacercarial stage has been suggested as a route of cerebral infection in animals^{15,16} and cannot be ruled out in this case.

The transparent corneal window afforded a unique opportunity to observe the movements of the fluke in human tissue. We observed its movement throughout the anterior chamber and watched it burrow through the iris to the posterior chamber. We observed its passage through the iris 7 times. Even in saline solution after its extraction from the anterior chamber the fluke continued to move in a fashion similar to that observed in vivo.

Intraocular paragonimiasis is very rare compared with other parasitic infections of the eye. The

diagnosis should be considered, however, when patients, especially children, from endemic areas present with repeated attacks of intraocular pain accompanied by uveitis, particularly when the inflammation is combined with multiple injuries to the eye. In most cases demonstration of ova in sputum and/or faeces and pulmonary involvement shown by chest x-ray are diagnostic. Intradermal or complement fixation tests are useful.

Treatment of intraocular paragonimiasis includes surgical extraction of the parasite as early as possible to arrest the symptoms and damage caused by migration of the fluke. When the worm invades the posterior chamber, vitrectomy has proved useful. Ocular steroids and mydriatics should be administered and systemic bithionol is essential.

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