

Intraocular pentastomiasis causing unilateral glaucoma

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SUMMARY We present a case of intraocular pentastomiasis in a 12-year-old Israel Arab boy. A single secondary pentastomid larva, most likely of *Linguatula serrata*, was found in the anterior chamber of the right eye, attached loosely to the pupil's border by a fibrinous mass. Associated conditions were iritis, subluxation of the lens, and secondary glaucoma. This is the first documentation of human pentastomiasis in Israel.

Cases of human pentastomiasis involving the eye are relatively rare. In so far as we have been able to trace them, there are only 10 previous reports of ocular pentastomiasis: four of these are from the USA and were due to larval *Linguatula serrata*¹⁻⁴; the remaining six are from Africa and were caused by *Armillifer armillatus*.⁵⁻¹⁰

The present case is of an Israeli Arab boy who suffered from unilateral glaucoma secondary to iritis which was provoked by a larval Linguatulid. This is the first report of human pentastomiasis from Israel.

Case report

A 12-year-old Israeli Arab boy from a neighbouring village was referred to our clinic because of high intraocular pressure in his right eye. The boy said he had suffered pain in his right eye for about a week, beginning after 'something believed to be a fly hit it'. On admission he had a visual acuity of 30 cm finger counting on the affected right eye. The intraocular pressure was 48 mmHg by applanation. The cornea was oedematous, with some flare in the anterior chamber. The pupil was 3 mm wide, round and free, the lens clear but with partial iridodonesis on the lower nasal quadrant. On ophthalmoscopy there was a glaucomatous cupping of 0.8 cup/disc ratio in the optic disc. Gonioscopy revealed several anterior goniosynechiae, particularly at 9 and 11 o'clock. The visual acuity of the left eye was 6/6, the intraocular

pressure was 12 mmHg, and the anterior and posterior segments were normal.

Secondary glaucoma of the right eye was diagnosed and the boy was given tablets of acetazolamide 0.125 g four times a day, topical drops of homatropine 1% with topical steroids three times a day and timolol maleate 0.25% twice daily. The following day, on mydriasis, a subluxation of the lens was observed. The intraocular pressure diminished to 28 mmHg but increased again to 35–40 mmHg on subsequent days. Nine days after admission, and on failure of the conservative treatment, an uneventful trabeculectomy was performed.

On the first postoperative day, in the course of slit-lamp examination, we noticed a white segmented mass in the anterior chamber which was encased in a fibrinous sheath and attached loosely to the pupil's border at 12 o'clock (Fig. 1). This fibrin coated and slightly crescent shaped mass changed its position several times during the next few days. Laboratory tests, including a complete haematological examination, gave normal results. Stool examinations for parasites were negative. Meanwhile there was no alleviation of the intraocular pressure.

One week after the trabeculectomy the mass was removed from the anterior chamber via limbal paracentesis. We were obliged to cut the fibrinous sheath in order to liberate the segmented mass and to take it out. After this procedure the eye became less irritated, and in the next few days the intraocular pressure decreased markedly, to stabilise at 13–14 mmHg.

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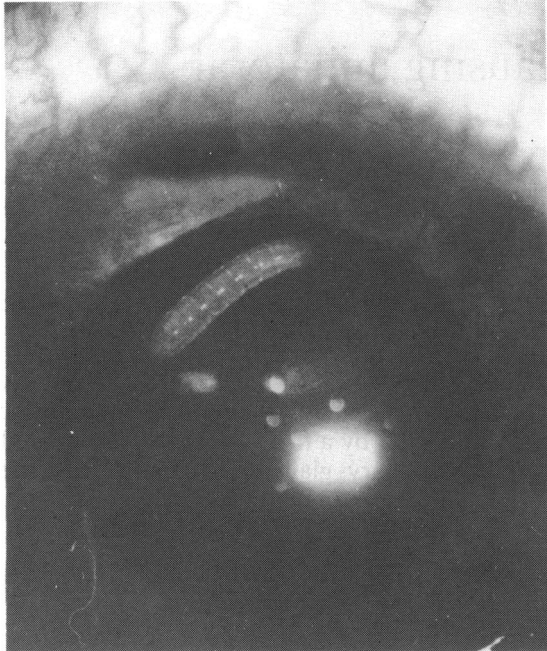


Fig. 1 Photograph (taken at a distance of 35 cm) showing slightly crescent shaped foreign body loosely attached to the right eye pupil at 12 o'clock. This was diagnosed as a second stage pentastomid larva, most likely of *Linguatula serrata*.

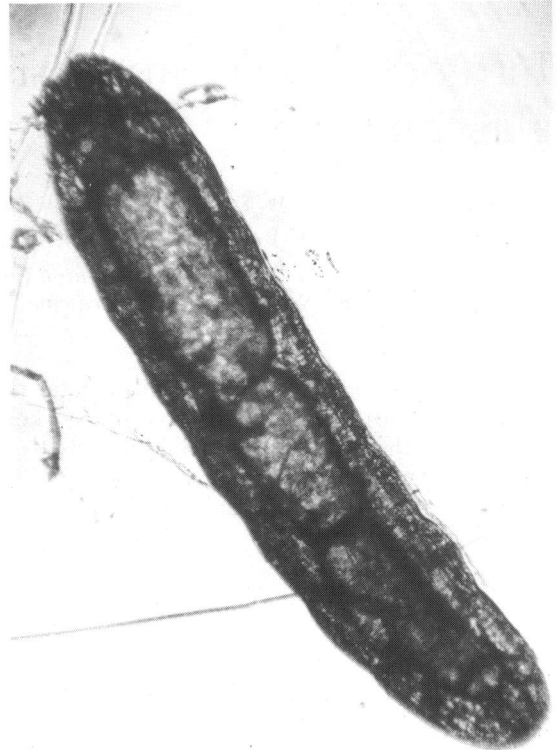


Fig. 2 The pentastomid larva after removal from its fibrous encasement. The specimen had undergone partial decomposition, but body cavity and openings at both ends were still discernible. The body of larva is only very sparsely annulated. $\times 9$.

Three weeks later the patient was discharged home. The visual acuity of the affected eye was then 1/2 a meter finger counting and the intraocular pressure was 10 mmHg. The anterior segment was normal apart from the existence of fibrin remnants in the anterior chamber, which were stretched over the lens and were attached to the limbal wound. On ophthalmoscopy the optic disc appeared pallid. The fellow eye seemed to be the same as on admission.

After he was discharged the boy was lost to follow-up for almost a year. When he was traced and examined at our outpatient clinic, there was no change in the intraocular pressure of the right eye, and the visual acuity was about the same as on discharge. However, a cataract had begun to develop, and the optic disc seemed to have undergone atrophy.

PARASITOLOGICAL FINDINGS

The extracted fibrinous mass had to be cut through in order to release the object contained inside. Once freed, the latter could be examined under the dissecting microscope and later cleared in 10% KOH. The object measured 2.3 mm in length and 0.5 mm in maximal width. It appeared to have undergone some decomposition within its fibrinous sheath, and at first

glance its cylindrical and slightly annulated shape was reminiscent of a fly larva (Fig. 2). However, closer examination after clearing failed to reveal the characteristic mouth hooks and posterior spiracles of the fly maggot, but instead showed the incipience of the anterior fang-like hooks typical of pentastomid infective stages (Fig. 3). Under higher magnification the fine spination of the cuticle was revealed (Fig. 4), and this, together with the size measurements, the absence of massive annulation of the body, and the geographic location, finally led us to identify the object as a secondary (second-stage) linguatulid larva (nymph), most likely of *Linguatula serrata*.

Discussion

The pentastomes or pentastomids are haematophagous metazoan endoparasites that as adults live in the lungs of reptiles and birds or in the nasopharynx of mammals, usually carnivores. The larval-nymphal

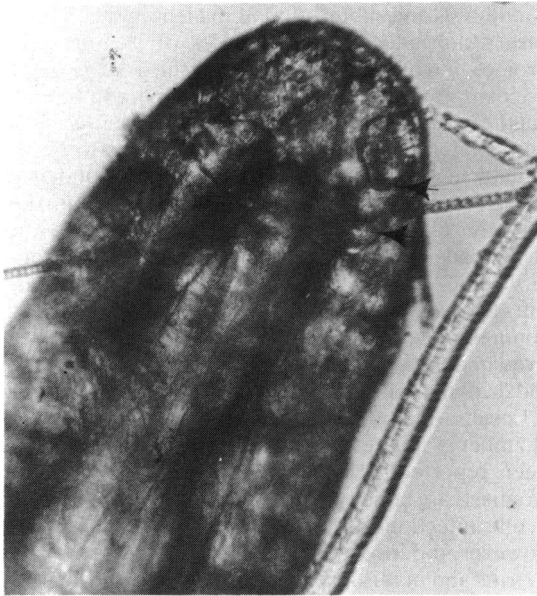


Fig. 3 Anterior end of pentastomid larva. Arrows mark incipient hooks on the one side. Spination of cuticle discernible at anterior margin on right. $\times 18$.

stages develop in a wide variety of vertebrate intermediate hosts, including man. Man, however, is usually an aberrant intermediate host, representing a dead end for the parasite.¹¹ The taxonomic position of pentastomes is still obscure, and exact details of the life cycles of most of the species are little known. The pentastomes seem to occupy an intermediate position between annelids and arthropods and have been assigned to a separate phylum (Pentastomida^{11,12}), but a more recent view¹³ is that

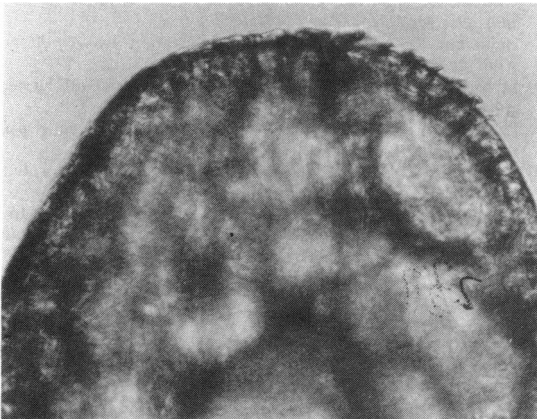


Fig. 4 Further enlargement of anterior extremity of pentastomid larva revealing the spination (visible on right side) exclusive to pentastomids of genus *Linguatula*. $\times 24$.

they are a subclass within the Crustacea. Only six species of pentastomes are known to infect man, and of these, two species, namely, *Armillifer armillatus* and *Linguatula serrata*, account for more than 99% of reported cases of human pentastomiasis.¹¹ The former is a species limited to Africa, while the latter has been recorded from man in Africa, Europe (Germany, Switzerland, Italy, Greece, and Turkey), the Near East and Middle East, and the American continent (Brazil, Chile, Colombia, Panama, Mexico, Cuba, and the USA).^{11,13}

The present intraocular case was diagnosed as due to a species of *Linguatula* and this for several reasons. Firstly, our patient has never travelled abroad. This rules out *Armillifer armillatus* and the related *A. grandis*, which are strictly African species. Secondly, the definitive hosts for *Armillifer* species are snakes of the genera *Python* and *Bitis*,¹⁴ and neither of these is represented in Israel. Thirdly, by its length (2.3 mm) the pentastomid removed by us occupies an intermediary position between the length ascribed by Hobmaier and Hobmaier¹⁵ for the second stage (0.35–0.5 mm) and third stage (4–6 mm) larva of *Linguatula rhinaria*, now regarded as a synonym of *Linguatula serrata*.¹⁶ Finally, and most importantly, of third stage pentastome larvae (nymphs) encountered in man all have a smooth cuticle except those of species of *Linguatula*, whose conspicuous spinules are sufficient to differentiate them from all the rest.¹¹ As Fig. 4 shows, our specimen bears the rudiments of such spination, although it still lacks the massive body annulation and the crescent or cashew nut shape of the infective, third stage larva.¹¹

As mentioned earlier, our patient has never travelled abroad, so clearly the infection was contracted locally. Ours is the first report of human pentastomiasis in Israel, and in view of the geographic location it is almost certain that we are dealing with a larval stage of the species *Linguatula serrata*. The adult stages of this species reside in the nasopharynx of carnivores, mainly dogs and cats, and these abound in Israel as well as in neighbouring Lebanon, where 44.3% of street dogs in Beirut have been found to harbour *L. serrata*.¹⁴

The life cycles of most species of pentastomes have not been adequately studied, but that of *L. serrata* is fairly well known thanks, inter alia, to Hobmaier and Hobmaier.¹⁵ Briefly, the developmental cycle requires carnivores (dogs and cats) as definitive hosts and herbivorous animals, mainly sheep and goats, as intermediate hosts. However, various other mammals, including man, can also serve as intermediate host. Eggs of the parasite, which are laid in the upper air passages of the definitive host, are passed to the exterior in its sputum or faeces, whence they are ingested by the intermediate host with

contaminated forage or water. The eggs hatch in the intestine, and the liberated first stage larvae, rarely exceeding 250 µm in length, penetrate the intestinal wall and migrate in the abdominal cavity to settle and ultimately encyst in various organs but mainly in the liver and lymph nodes. After several moults within the cyst, the third stage larva (also called nymph or juvenile) emerges from its cyst to migrate again through the peritoneal cavity before re-encysting in various tissues and organs. When these are now consumed by the carnivorous definitive host, the infective larva excysts to travel via the stomach and oesophagus to the nasopharynx, where, after several additional moults, it attains sexual maturity.^{11 13 14}

Man—an aberrant intermediate host—usually acquires the infection from egg contaminated vegetables or water, or perhaps from intimate contact with an infected cat or dog,¹¹ but also can become infected by consuming the raw liver and/or lymph nodes of sheep and goats which contain the encysted tertiary linguatulid larvae. The latter form of human pentastomiasis, characterised by severe nasopharyngitis, is called the halzoun or marrara syndrome,¹⁷ and is prevalent in Lebanon, Turkey, and Greece.¹¹

The present and similar cases of ocular pentastomiasis pose the interesting question as to how the parasite reaches this organ. The question has intrigued several investigators¹⁸ and has yet to be resolved. In the present instance the patient claimed to have been hit in the eye by a fly a week prior to his admission. Some flies, such as *Oestrus ovis*, do infest the human eye with their maggots, producing ophthalmomyiasis, but there is no record as yet of flies serving as vectors of pentastomes, though there has been one report of a cockroach (*Periplaneta americana*) serving as intermediate host for a lizard pentastome.¹⁹ Moreover, the finding of a unilateral glaucomatous excavation of the optic disc and the presence of several goniosynechiae in the boy's right eye strongly suggest a parasitic infection of a much longer duration than one week.

From the life cycle of pentastomes, as briefly described above, it is clear that both the first stage larva (emerging from the egg) and the third stage larva (after excystment) have a propensity for migration in the body of the host. Might not the microscopic first stage larva, which rarely exceeds 250 µm in length, inadvertently enter a blood vessel, in the course of its penetration through the intestinal wall, and be transported to the eye? We believe this is at least theoretically feasible, and other parasites, such as *Entamoeba histolytica*, do occasionally travel in the blood stream to reach the human brain. Since the migratory pentastome stages are endowed with proteolytic capabilities, these could explain the observed

zonulolysis and subluxation of the lens in the present case. Alternatively, in the cases of halzoun syndrome, might not the liberated third stage larva accidentally travel from the nasopharynx to the eye via the nasolacrimal duct, which is a sufficiently wide, non-ciliated passageway? We believe this to be an equally feasible explanation for some cases of ocular involvement due to *Linguatula serrata*, but in our case the first conjecture is preferable because the recovered parasite was apparently a second stage larva and there was no history of halzoun. The infection could readily have been acquired from egg contaminated vegetation or water, because the boy lived under very poor sanitary conditions and admitted drinking water from doubtful sources.

Localisation of pentastomid larvae in the anterior chamber is apparently rare, for only a few cases have been reported.^{1-3,9} Ocular manifestations of pentastomiasis are equally rare, and indeed Fain¹⁶ refers to ocular infections as being free of lesions. Our case, however, did involve unilateral glaucoma secondary to iritis and in this respect and also in its aetiology, is almost identical to that reported by Deweese *et al.*³ from a Negro girl.

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