

INTRA-OCULAR COENURUS INFESTATION

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Coenurus is the term applied to the cystic larval stage of a tapeworm in which there are numerous scolices in contrast with cysticercus, and no evidence of daughter cysts as in hydatid. This particular larval form, as well as the characteristic shape of the rostellum and hooklets and certain features of the vagina in the adult worm, are considered diagnostic systematic traits of the subgenus *Multiceps*.

The proper host of the adult worm of a number of species of *Multiceps* is not known, but it is safe to say that various carnivores, especially canines, harbour it. Intermediate hosts may vary greatly. The greatest number are to be found among the rodents, but various primates have been incriminated who, it may well be, are incidental intermediate hosts, having been infested along the same lines as man, in whom sporadic cases have been reported.

Multiceps coenurus (*coenurus cerebralis*) has a wide host distribution. It varies morphologically according to the intermediate host and the tissue in which it is found. Though it is most frequently reported as inhabiting the central nervous system, *coenurus* occurs in animals in the intermuscular connective tissue, the abdominal cavity, the thoracic sheath, and the pericardial sheath. Many human *coenurus* infestations are doubtlessly passed off as hydatid cysts.

The following case, which illustrates the value of a surgical exploration of the uveal tract, is only the third case on record of an intra-ocular *coenurus* infestation.

The first case was reported by Boase¹ and by Raper and Dockeray² in an adult Native male in Uganda. Clinically, a large whitish cyst was noted projecting from the nasal side of the fundus, apparently filling the vitreous and in contact with the posterior surface of the lens. Movements were detectable in the cyst, suggesting two scolices. The eyes was very painful and the patient requested enucleation. He had presented himself at the clinic 10 months earlier with signs and symptoms of iritis. Atropine drops were instilled into the eye and he was asked to wait for further examination, but unfortunately did not do so. Examination of the enucleated eye revealed a trilobular cyst with numerous scolices and was considered to be *Multiceps coenurus*.

The second case was reported by Wainwright,³ of Durban, in an enucleated eye. A Bantu male aged 51 complained of a painful eye; the condition was diagnosed as conjunctivitis and iritis. Unfortunately no ophthalmoscopic report is available. Four months later there was a marked increase in tension, great pain, proptosis, ciliary staphyloma, and dense keratic precipitates. The eye was enucleated with a tentative diagnosis of neoplasm. Examination revealed a bilobular cyst filling the vitreous, which was identified as *coenurus cerebralis*.

PRESENT CASE

The present case concerned a female European child aged 3½ years. She was born in the Transvaal but for the year before consultation had been resident in Swaziland. The mother had noticed that during the previous 2-3 months the left eye had been variably congested and that at times the child complained that it was sore. The mother thought that the child had bumped the eye when the symptoms first appeared; she imagined that she saw a bruise just lateral to the eye. The patient's doctors had treated her for conjunctivitis.

Examination showed a mildly flushed eye, a small clotted streak of blood on the iris, and a few delicate posterior synechiae. The vitreous was very hazy and fundal details could not be seen, but it was obvious that a white raised mass was present on the nasal side. The tension of the eye was normal.

Good and complete mydriasis was obtained with homatropine and cocaine drops plus one drop of 1% adrenalin. It was then obvious that an extensive smooth high elevation of the nasal fundus was present, reaching from the ciliary body to apparently the region of the disc, although it was extremely difficult to define the disc on account of the vitreous haze. The maximal elevation was just below the horizontal meridian between the ora serrata and the equator. The swelling tapered off below and above, so that the retina appeared flat in the oblique meridians although it had the same oedematous whitish appearance as on the swelling itself. No movement was noticed in the mass, which was specially examined for this phenomenon. The temporal half of the retina appeared normal, but the anterior region only could be examined through the cloudy vitreous.

As far as could be ascertained in a child of this age there was no perception of light with this eye. Transillumination under general anaesthesia showed the mass to be translucent.

The findings suggested a granulomatous uveal inflammatory lesion and, amongst the possible causes, tuberculosis, Coats's disease, cysticercosis, hydatid disease and even hookworm infestation were considered. Neoplastic disease was excluded as unlikely on the physical findings. This is discussed further below.

Investigations

Haemoglobin 14.5 g%. Leucocytes 9,600 per c. mm. (neutrophils 52%, lymphocytes 40%, monocytes 4%, eosinophils 3%, basophils 1%). Sedimentation rate, 17 mm./hour. Ide test negative.

Stool examination for parasites, negative. Hydatid complement-fixation test, positive (result received after operation).

X-ray examination of head, chest and legs showed no abnormality.

Operation

Surgical exploration of the mass was considered at least for biopsy to pave the way if possible for rational therapy. Under general anaesthesia a large scleral flap was reflected. The two parallel incisions starting a few mm. from the limbus were 2-3 mm. above and 4-5 mm. below the borders of the medial rectus muscle. A vertical incision joining these two horizontal cuts was placed a few mm. behind the insertion of the medial rectus, which

muscle had first been detached. The larger posterior flap extended to just behind the equator, the furthest point that could be reached without undue traction on the globe. Greater details of a similar approach for removal of a cysticercus have been published elsewhere.⁴ The eye had been softened as much as possible by pre-operative 'diamox' retrobulbar anaesthesia, and finally a paracentesis of the anterior chamber just before the scleral incisions were carried through to the choroid. The exposed choroid was paler than normal. The region corresponding with the maximal swelling in the fundus was gently incised, and suddenly a small white cystic mass herniated through the incision. A single scolex was visible in the small bladder and the whole simulated a typical cysticercus cellulosae. With this assumption in mind a pair of iris forceps were applied with closed blades to sweep the cyst from the wound in the choroid. The gentle pressure applied made the cyst bulge, and more scolices became evident. The choroidal wound was then enlarged and the endocyst of what was then thought to be a hydatid oozed out with a little pressure. Part of the outer ectocyst wall was excised for investigation and the remaining now collapsed cavity was gently swabbed with 10% formalin. The cavity extended from about the middle of the ciliary body to midway between equator and disc.

The adjacent tissue was then examined, both externally and by means of the headlamp ophthalmoscope worn throughout the operation,⁵ to exclude the chance of a second cyst. Six 0 mild chromic catgut was used for closure of all incisions and reattachment of the medial rectus muscle. Diathermy was available to coagulate any raw surface should a biopsy only have been taken, e.g. in the case of a tuberculoma. A long-acting antibiotic preparation was given once postoperatively and oral steroid therapy was also commenced. Convalescence was uneventful.

The vitreous cleared and the oedema of the nasal retina settled, but its appearance was abnormal, suggesting a gliosis and fibrosis with patchy pigmentation. The temporal region appeared normal. The optic disc was very distorted, especially its nasal side. Four months after the operation the disc was still red, but there was apparently no return of vision, although the pupil reacted a little to light. Cosmetically, however, the eye appeared normal. Unfortunately follow-up was limited to 6 months, when contact was lost.

Pathological Findings

The cyst was about 8 mm. in diameter but of rather irregular shape (Fig. 1). Numerous white nodules of about 1-2 mm. in diameter were clearly visible in the wall of the cyst; fresh preparations showed these to be definite scolices (Fig. 2). The individual scolices consisted of a rostellum with a double row of hooklets and 4 suckers typical of the taenia type of tapeworm (Fig. 3). There were no daughter cysts. The hooklets of the armed rostellum could not be measured in the original condition, so that it was necessary to tease one of the scolices apart. There were 2 rows of hooklets present. Measurements of both large and small hooks showed that they clearly fell within the small variation limit of *Multiceps multiceps*—large hooks 150-170 μ and small hooks 90-130 μ as given by Baer.⁶ Hooks are known to be reliable morphological features because numbers, sizes and shapes are rather constant for each species.

DISCUSSION

Human infestation by cysticercus cellulosae, coenurus cerebri and hydatid is known to occur in South Africa. The brain, meninges and cerebrospinal-fluid pathways are not infrequently involved, with serious results.^{3,7-12}

This is the second case of *Multiceps coenurus* in the eye described in South Africa. It is probable that this child was infected from the ingestion of eggs passed in the stools of an infected dog. However, attempts to trace infection to the child's pet dog have proved fruitless, so that the source of the infection remains unknown.

Coenurus cysts appear to develop in man in certain well defined situations only; these include the cerebrospinal-fluid pathway and, as indicated by this case, the uvea of the eye. Degenerated cysts are sometimes encountered in the central

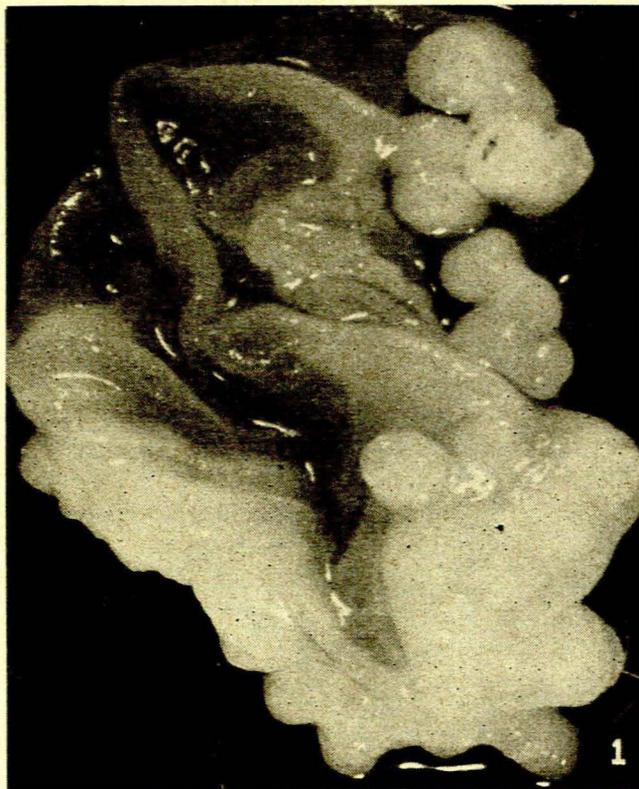


Fig. 1. Macroscopic appearance of the opened fresh cyst, $\times 12$.

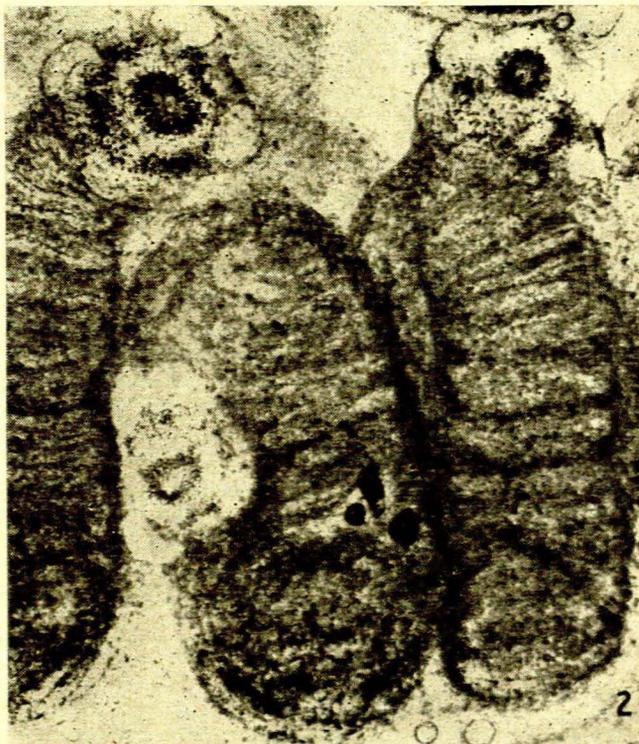


Fig. 2. Scolices evaginated from the wall of the cyst, $\times 38$.

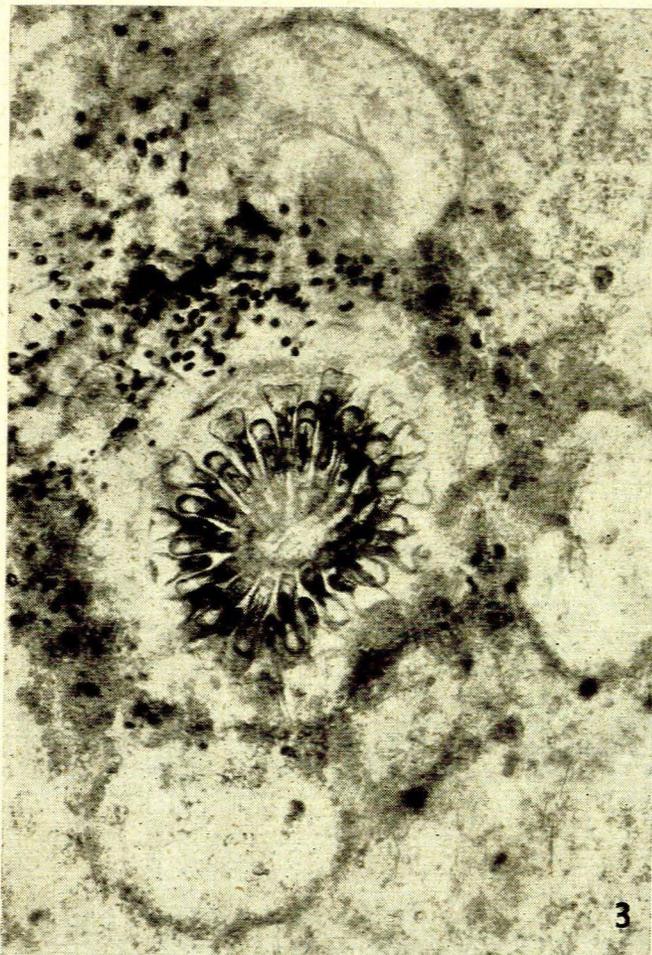


Fig. 3. The rostellum of one of the scolices, showing 3 of the 4 suckers and the double row of hooklets, $\times 150$.

nervous system which are obviously derived from tapeworms, but their true identity cannot be determined because of the absence of scolices. Similar cysts, containing numerous scolices, have been described in the Belgian Congo, where Fain has succeeded in identifying them as the larval stage of the tapeworm *Multiceps (Taenia) braunii*.¹³

Intra-ocular helminthic infestations occur rarely. Cestodes, nematodes and diptera are recorded. Hydatid cysts within the eye are very rare. Cysticerci are more frequent. When the parasite has ruptured into the vitreous, the diagnosis is fairly simple. However, when the cyst is subretinal and especially when it is within the uvea, the diagnosis becomes more difficult. The problem of malignant neoplasm then arises.

In the adult a melanoma might present the greatest difficulty. Parasitic cysts usually produce signs of inflammation, but so may the melanomas, especially when

undergoing necrosis.¹⁴ Melanomas, however, usually occur as large tumours, which makes the diagnosis almost self-evident, whereas a small cyst with inflammatory changes is more likely a parasite. The features of pigmentation and transillumination help, but cystic change in the necrotic centre of a melanoma might make differentiation almost impossible.

In the young child a malignant melanoma is exceptional¹⁵ and one completely translucent with inflammatory changes probably hypothetical. The more likely problem would be the exophytum type of retinoblastoma. This would present with a smooth elevated retinal surface, but clinical inflammatory changes have not been described with this type of tumour.¹⁴ Wilder¹⁶ described 46 enucleated eyes from young children where a clinical diagnosis of retinoblastoma had been made and in which histologically no tumours were found. The pathological diagnosis of pseudo-tumour or Coats's disease was made. Re-examination of these 46 eyes by serial sections then showed the definite presence of nematode larvae in 24 and bodies suggestive of degenerating larvae in the remaining 22. Although it was impossible to determine the exact genus of the larvae they were considered to be hookworm.

Probably the earliest features in the case described in this paper, and in the 2 previously described cases in Natives, were a localized swelling in the fundus, vitreous haze, and an iritis. It is suggested that cases with such features—selected after due consideration of laboratory investigations, increasing swelling and inflammatory reaction, etc.—should be submitted to what might be called exploratory sclerotomy. Had this case been seen sooner there is every chance that useful vision would have been saved.

SUMMARY

A case of intra-ocular infestation with a coenurus of *Multiceps multiceps*, and its successful removal, is described. Aspects of intra-ocular helminthic infestation and the relation to the diagnosis of pseudo-tumour are discussed. The value of exploratory sclerotomy is suggested.

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