Japan. J. Med. Sci. Biol., 38, 223-227, 1985.

Short Communication

A REPORT OF THE FIRST CASE OF *DIROFILARIA* INFECTION IN THE EYELID REGION IN JAPAN

Noboru KAGEI, Kengo TANAKA¹, Ryoichi OKAMURA¹, Masanori KORENAGA² and Isao TADA²

Department of Parasitology, National Institute of Health, Kamiosaki, Shinagawa-ku, Tokyo 141, ¹Department of Ophthalmology, and ²Department of Parasitic Diseases, Kumamoto University School of Medicine, Kumamoto 860

(Received October 15, 1985. Accepted December 3, 1985)

SUMMARY: *Dirofilaria* worms were recovered from a painless hard tumor in the right eyelid of a 19-year-old Japanese male who had lived in Kagoshima and Kumamoto Prefectures, Japan. In the worms, the cuticle had a smooth surface, the inner cuticular layer had internal lateral longitudinal ridges, and the musculature of the body wall was the polymyarian type. On the basis of all morphological characteristics observed, this worm was identified as an immature female *Dirofilaria immitis*. This is the first human case of nematode infection in the eyelid in Japan. Surgical removal of the mass with worms resulted in complete recovery from swelling of his right eyelid.

Dirofilaria spp. are widely distributed and cases of human dirofilariasis are often observed in the world. Many reports have been published on the disease, while there have been only occasional reports in the field of ophthalmology. Although about 30 cases of human dirofilariasis have been reported in Japan, *Dirofilaria* worms have been found in the lung or the pleural cavity and subcutaneous tissue in most cases. No case of human ocular dirofilariasis has been recorded in Japan. In this report, the authors present a case of *Dirofilaria* infection in the eyelid that caused symptoms of inflammatory pseudotumor.

The patient was a 19-year-old Japanese male student who was born in Kagoshima Prefecture, Japan, and lived there ever since. Recently, he has moved to Kumamoto Prefecture. The patient complained of swelling and redness of the eyelid and conjunctival hyperemia of the right eye which had begun two days before. There were no pain or itching in the lesion. He consulted an ophthalmologist on October 24, 1983. On the examination, visual acuities of both eyes proved to be normal. The disease was provisionally diagnosed by the doctor as external hordeolum in the right upper lid. Although he was orally administered some drugs and received eye wash, the exact names of the drugs were not learnt because he has been unable to contact the doctor after that. Three weeks later, when the mass lesion was essentially unchanged

影井 昇 (国立予衛生研究所寄生虫部)

田中謙剛・岡村良一 (熊本大学医学部眼科学教室 熊本市本荘 2-2-1)

是永正敬・多田 功 (熊本大学医学部寄生虫病学教室 熊本市本荘 2-2-1)

despite of the treatment, he visited the Department of Ophthalmology, Kumamoto University School of Medicine.

Although the result of general physical examination was not special, the eye palpation revealed a subcutaneous painless firm mass measuring 2.5 cm by 1.5 cm in the right upper lid. The mass lesion with the well-defined margin was confirmed also by computerized tomography scanning. Under the diagnosis of tumor, an incision was performed on the right upper lid and the affected part was removed. The swelling of the eyelid disappeared immediately after the surgical treatment.

The extirpated lump was fixed with formalin and sectioned for staining. Three or four fragments of a nematode were observed in the central region of each abscess and necrosis was also found in the tissue between such fragments (Fig. 1). The giant epithelioid cells and macrophages were found in the surroundings of the worm (Fig. 3). The abscess adjacent to the worm fragments consisted of inflammatory granuloma which was characterized by the existence of newly born blood vessels, fibroblasts, macrophages, neutrophils, lymphocytes and many eosinophils.

Although the serum was immunologically examined by the Ouchterlony and immunoelectrophoresis techniques using *Ascaris lumbricoides*, *Toxocara canis*, *Anisakis simplex*, *Dirofilaria immitis*, *Parascaris equorum*, *Fasciola hepatica*, *Paramphistomum cervi*, *Dipylidium caninum* and *Taenia saginata* as antigens, the results were negative to all antigens except *T. canis*.

The leukocyte count on the peripheral blood smear was 7,000 per μl with differential counts of staff cells 12%, polymorphonuclear leukocytes 61%, lymphocytes 26%, eosinophilis 1%, monocytes 1% and basophilis 1%. The erythrocyte count was 504×10^4 per μl . The packed cell volume by hematocrit was 48%, hemoglobin 15.1 g/dl and the thrombocyte count 2.54×10^4 per μl . The blood was negative for microfilariae by the direct smear technique.

Under the microscope, several oblique and transverse sections of the worm were seen in the pathological specimens cut at various levels (Fig. 1). The distances between fragments were 260 to 928 μm . The 48 cross sections of the worm measured 47 to 124 μm in width. Some portions of the worm showed degenerative changes in which only the cuticle and muscle fibers of the body wall were distinguishable from other degenerated tissues. The thickness of the cuticle ranged from 4 to 7 μm , depending on the location in the worm. The outer surface of the cuticle was smooth and provided with neither ridge nor projection in the transverse sections, but diagonal fibrous layers of the cuticle were seen in the tangential section (Fig. 2). An internal longitudinal cuticular ridge was clearly recognizable in each innermost portion of a pair of lateral chords in transverse sections (Fig. 3). The muscle cell arrangement was of polymyarian type. The fiber zone of the muscle was 20 μm thick. In some fragments of the nematode, there were two or three uteri in the center (Fig.

Fig. 1. Four transverse sections of the worm found in an eye nodule (hematoxylin-eosin stain). x67.

Fig. 2. Diagonal fibrous layers of the cuticle seen in a tangential section of a worm (silver stain). x650.

Fig. 3. A transverse section of a worm showing an internal longitudinal cuticular ridge (arrow) in each innermost portion of a pair of lateral chords. The giant epithelioid cells and macrophages were found in the area surrounding the worm (hematoxylin-eosin stain). x325.

Fig. 4. A transverse section of the worm showing three uteri in the center of a fragment (hematoxylin-eosin stain). x325.

4). These uteri measured 45 to 57 μm in length and 27 to 30 μm in width. In some fragments, the uteri seemed to contain many primary oocytes, showing that the worm was female, but had no microfilaria.

Faust (4) compiled a useful list of cases of human infection with *Dirofilaria* spp. Since then, many human cases have been reported by many workers in various geographic sites of the world.

Although about 43 cases of human ocular dirofilariasis have been reported in the world, almost all *Dirofilaria* worms recovered from the eyes were identified as *D. conjunctivae* and *D. repens*. Only Dissanaïke et al. (3) reported a case of immature *D. immitis* infection in the vitreal body of man in Malaysia, and Christie (2) on a case of *D. tenuis* infection in a human eye in Holland.

Adult *Dirofilaria* worms found in Japan have been identified as *D. immitis*, *D. repens* or *D. ursi*. As shown by Yoneda et al. (13), *D. immitis* is a very common parasite of the dog in the southwestern district of Japan (63% in pound dogs), whereas *D. repens* has been found only in Okinawa prefecture (7) and *D. ursi* is reported only in the central district of this country (11,12).

In the present case, we identified the worm as an immature female of *D. immitis*, by the following characteristic features; the smooth outer surface of cuticle, internal longitudinal cuticular ridges and polymyarian musculature (1,5,6,8-10). Although the Ouchterlony and immunoelectrophoresis techniques using adult *D. immitis* as the antigen gave no plausible result on immunodiagnosis, this does not rule out the morphological conclusion. The reason for the negative results on such serological studies is not clear.

Because of the migratory habit of the larvae in various tissues and of various symptoms induced there, human dirofilariasis should be taken hereafter into consideration in many fields of clinical medicine.

ACKNOWLEDGEMENTS

We are grateful to Professor M. Tsuji, Hiroshima University School of Medicine, for his help in serological examinations.

REFERENCES

1. Billups, J., Schenken, J. R. and Beaver, P. C. (1980): Arch. Pathol. Lab. Med., 104, 11-13.
2. Christie, R. W. (1977): New Engl. J. Med., 297, 706-707.
3. Dissanaïke, A. S., Ramalingam, S., Fong, A., Pathmayokan, S., Thomas, V. and Kan, S. P. (1977): Amer. J. Trop. Med. Hyg., 26, 1143-1147.
4. Faust, E. C. (1957): Z. Tropenmed. Parasitol., 8, 59-68.
5. Font, R. L., Neafie, R. C. and Perry, H. D. (1980): Arch. Ophthalmol., 98, 1079-1082.
6. Gutierrez, Y. (1984): Human Pathol., 15, 514-525.

7. MacLean, J. D., Beaver, P. C. and Michalek, H. (1979): Amer. J. Trop. Med. Hyg., 28, 45-48.
8. Neafie, R. C., Connor, D. H. and Meyer, W. M. (1976): *Dirofilariasis. Pathology of Tropical and Extraordinary Diseases.* C. H. Binford and D. H. Conner [eds.], Vol. 2, 391-396. Forces Institute of Pathology, Washington.
9. Ohmori, Y., Yamaguchi, T. and Ohishi, I. (1977): Japan. J. Parasitol., 26 323-335 (text in Japanese).
10. Orihel, T. C. (1961): J. Parasitol., 47, 251-262.
11. Uni, S. (1978): J. Osaka City Med. Center, 27, 439-458 (text in Japanese).
12. Yamaguti, S. (1941): Japan. J. Zool., 9, 409-439.
13. Yoneda, Y., Hirota, M. and Takao, Y. (1979): J. Kurume Med. Assoc., 42, 1020-1025 (text in Japanese).