SECOND CASE OF ZONOTIC *ONCHOCERCA* INFECTION IN A RESIDENT OF OITA IN JAPAN

TAKAOKA H.*, BAIN O.**, TAJIMI S.***, KASHIMA K.****, NAKAYAMA I.****,
KORENAGA M.*****, AOKI C.* & OTSUKA Y.*

Summary :

A non-gravid female *Onchocerca* was found in histopathological sections of a biopsy specimen taken from a painful nodule in the wrist of a 57-year-old woman in Oita, in southern Japan. Six species of *Onchocerca* have been found in animals in Japan: two in wild bovines, one in equids, and three in domestic bovines of which one, *Onchocerca* sp., is only known by the microfilaria and infective stage. Distinctive morphological features of the worm, including a three-layered thick cuticle with prominent annular ridges at wide intervals, high somatic muscles and narrow lateral chords, resembled those of *O. gutturosa*, one of the three bovine *Onchocerca* species transmitted in the Oita region. However *Onchocerca* sp., which is also transmitted in this region, cannot be excluded.

An ELISA test of the patient serum suggests that infections by *Onchocerca* spp. might be distinguished from those by *Dirofilaria immitis*, of which the number of human cases is increasing in Japan.

KEY WORDS : *Onchocerca*, zoonosis, Japan.

Résumé : DEUXIÈME CAS D'INFECTION D'UN HABITANT D'OÏTA, AU JAPON, PAR UNE ONCHOCERQUE ANIMALE

A Oïta, au sud du Japon, une femme consulte pour un nodule sous-cutané douloureux au poignet accompagné d'une paralysie récente de la main. Une onchocerce femelle, non gravide, est trouvée sur les coupes histologiques de la biopsie.

Six espèces d'onchocercques sont signalées au Japon: deux chez un bovidé sauvage en montagne, une chez les chevaux, trois chez les bovins domestiques dont une, *Onchocerca* sp., connue seulement par la microfilaire et le stade infectant.

Les caractères morphologiques distinctifs (cuticule épaisse à trois couches et aux côtes très saillantes et espacées, cellules musculaires hautes et étroites, cordes latérales étroites) ressemblent à ceux d'*O. gutturosa*, une des trois espèces bovines transmises à Oïta. Cependant *Onchocerca* sp., également transmis dans la région, ne peut être exclu.

Un test ELISA du sérum de la patiente suggère que les infections par *Onchocerca* spp. peuvent être distinguées des infections par *Dirofilaria immitis*, dont la fréquence augmente au Japon.

MOTS CLÉS : *Onchocerca*, zoonosis, Japan.

INTRODUCTION

Zoonotic onchocerciasis is very rare in humans. Only five cases have been reported, one each from the Crimea (Azarova *et al.*, 1965), Switzerland (Siegenthaler and Gubler, 1965), Canada (Ali-Khan, 1977), the USA (Beaver *et al.*, 1974) and Japan. The Japanese case was that of a 2-year-old girl living in Oita who developed a subcutaneous swelling on the sole of her foot, in which a female *Onchocerca* was found (Beaver *et al.*, 1989; Hashimoto *et al.*, 1990). A similar case, which was again found in a Japanese resident of Oita, is reported here.

* Division of Medical Zoology, Oita Medical University, Hasama, Oita, 879-55, Japan.

** Biologie Parasitaire, Protistologie, Helminthologie, CNRS URA 114, Museum National d'Histoire Naturelle, 61, rue Buffon, 75231 Paris, Cedex 05, France.

*** Musashi Orthopedic Clinic, Musashi, Higashikunisaki, Oita, 873-14, Japan.

**** Department of Pathology, Oita Medical University, Hasama, Oita, 879-55, Japan.

***** Department of Parasitology, Kochi Medical School, Nankoku, Kochi, 781-51, Japan.

Correspondence: Hiroyuki Takaoka. Tel. : +81.975.86.5701 – Fax : +81.975.86.5118 – Email:takaoka@oita-med.ac.jp.

CASE REPORT

A 57-year-old woman living in the Kunisaki district in Oita in southern Japan, developed a subcutaneous swelling on the left wrist (Fig. 1). It had been present for three years, and no symptoms were evident until she felt pain and her hand became paralysed. A biopsy specimen, which was taken from the basal part of the nodule measuring 4.5 cm long by 1.0 cm wide, was sectioned and stained in hematoxylin-eosin.

Six sections available for study showed the presence of a non-gravid female adult filaria, which was assigned to the genus *Onchocerca*, based on its morphological characters. The worm was present in the deep dermis adjacent to the subcutaneous tissues. It was completely surrounded by epithelioid granulomatous tissues comprised of an inner layer of polygonal or flattened multinucleated giant cells and several layers of epithelioid mononuclear cells. Infiltrated lymphocytes and plasma cells, intermingled with numerous eosinophils, were seen mainly around the granulomatous lesions and sporadically here and there even in the subcutaneous tissues. A moderate amount of fibrous

Fig. 1. – Swelling of the wrist at the level of the tumor (arrow).

Figs 2 and 3. – Histological sections of the worm and tissular reaction.

tissues was developed in the dermis and subcutaneous fat tissue.

The description of the worm was based upon histological sections and upon a small fragment recovered at dissection of the remnants of the nodule (Figs 2, 3 and 4). The cuticle was trilaminar with prominent transverse straight ridges. The hypodermis was very thin in the submedian fields and thickened in the narrow lateral chords. The somatic muscles were well developed and of polymyarian-coelomyarian type. The number of muscle cells was difficult to count but appeared to vary from 7 to 12 per quadrant according to the diameter of the sections. The intestine was very small. The two uterine branches were empty.

Measurements were: diameter of the body from 182 to 260 μm ; cuticle 30 μm thick; width of a ridge 22 μm ; space between two ridges 67 to 105 μm ; muscle cells 30 μm high; intestine 15–22 μm wide.

Serum taken from the patient was immunologically tested by ELISA (Korenaga *et al.*, 1983) against antigens of *O. gutturosa*, *Dirofilaria immitis*, *Trichinella spiralis* and *Strongyloides ratti*. Sera positive and negative, for *O. volvulus*, each obtained from a Guatemalan and a Japanese, were used as controls for the test against the *O. gutturosa* antigen. The results show that the patient's serum was exclusively positive with an antigen of *O. gutturosa* (Fig. 5).

DISCUSSION

Six *Onchocerca* species have been reported in Japan (Sato *et al.*, 1954; Takaoka, 1990; Takaoka and Bain, 1990; Yagi, Bain and Shoho, 1994): three parasitic in bovines, *O. lienalis* Stiles, 1892, *O.*

Fig. 4. – Morphology of the worm; A: transversal histological section; B: lateral view of the female fragment recovered (scale: A, 100 μm ; B, 200 μm).

Fig. 5. – The ELISA values of the patients serum against *Trichinella spiralis*, *Strongyloides ratti*, *Dirofilaria immitis*, and *Onchocerca gutturosa* antigens. Y. K., patient's serum; negative control, Japanese pooled sera; positive control, Guatemalan pooled sera of patients with onchocerciasis.

gutturosa Neumann, 1910 and *Onchocerca* sp. sensu Takaoka et Bain, 1990, only known by the microfilaria and the infective stage, one parasitic in horses, *O. cervicalis* Railliet and Henry, 1910 and two parasitic in *Capricornis crispus*, *O. suzukii* Yagi, Bain and Shoho, 1994 and *O. skrjabini* Ruklyadev, 1964.

In the identification of the present specimen, three of these species can be excluded on morphological criteria: *O. suzukii* by the absence of ridges, *O. cervicalis* by ridges much closer together (16–25 μm instead of 105 μm), *O. lienalis* by less prominent ridges (Bain *et al.*, 1978; Eberhard, 1979).

On the other hand, the cuticle, the narrow hypodermic lateral chords and the well-developed muscle cells of this material resemble those of *O. skrjabini* (cf. Bain and Schulz-Key, 1974) and *O. gutturosa*. The presence of the former species in humans is improbable: it has not been recorded in Oita and the host exists in mountainous areas of Mt. Sobo in southern Oita. The patient was a resident of Kunisaki in northeastern Oita, and suspected of having become infected with this filaria in or near her living area. The cuticular ridges of *O. gutturosa* are often sinuous, but portions of the body present straight ridges, as drawn by Neumann (1910).

Thus, our worm is likely to be *O. gutturosa*, although *Onchocerca* sp., which is also frequently found in the Oita region, cannot be excluded.

Both species were reported to be transmitted by a simuliid, *Simulium bidentatum* in Oita (Takaoka,

1994). Being not only bovipilic but also anthrophilic, this black fly species may be a probable transmitter of these bovine *Onchocerca* to human. Our immunological examination could not identify the parasite precisely. But it suggests that human infection by *Onchocerca* spp. might be distinguished by an ELISA method from those by *D. immitis*, of which number of human cases is increasing in Japan (Makiya, 1990).

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