SECOND CASE OF ZOONOTIC ONCHOCERCAL 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 bovids, one in equids, and three in domestic bovids 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 EUSA 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.

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.

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 trilaminate 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. 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, 1996).
tissue pathology and comparative microanatomy of Onchocerca from a resident of Ontario and other enzootic Onchocerca species from Canada and the USA. Annals of Tropical Medicine and Parasitology, 1977, 71, 469-482.


NEUMANN L. G. Un nouveau Nématode parasite du Bœuf (Onchocerca gutturosa. sp.). Revue vétérinaire, 1910, 5, 270-278.


TAKAOka H. Natural vectors of three bovine Onchocerca species (Nematoda: Onchocercidae) and seasonal transmission by three blackfly species (Diptera: Simuliidae) in central Kyushu, Japan. Journal of Medical Entomology, 1994, 31, 404-416.


Reçu le 22 février 1996
Accepté le 19 mars 1996