Case report

A new case of zoonotic onchocercosis in northern Kyushu, Japan

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ABSTRACT

A case of zoonotic onchocercosis has been found in a resident who lived in Iizuka City, Fukuoka Prefecture, Japan for some time. A 24-year-old male developed a painful nodule on the middle finger of his right hand. The nodule was surgically removed from the vagina fibrosa tendinis of the finger at Beppu Medical Center, Beppu City, Oita Prefecture in 2012. The causative agent was identified as a female Onchocerca dewittei japonica based on its histopathological characteristics. The identity of the filaroid has been confirmed by sequencing the coxl gene. The present study indicates that the zoonotic onchocercosis caused by O. dewittei japonica has been concentrated in northeast Kyushu.

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Zoonotic Onchocerca infections have recently increased worldwide. Global warming, deforestation, and human demographics have affected the transmission of parasites among vectors, host animals, and humans [1]. These factors resulted in more concentrated occurrence of vector-borne parasitic zoonoses in areas where such infections have been previously reported. To date 31 cases of zoonotic onchocercosis have been reported in the world and seven Onchocerca species (Onchocerca gutturosa, Onchocerca cervicalis, Onchocerca dewittei japonica, Onchocerca jakutensis, and Onchocerca lupi) have been identified as the causative agents [2–5]. For example, since the first description of a human case of infection by O. lupi in 2011 [2], up to eight human cases have been reported so far, in countries in the Mediterranean basin and in the United States of America. In Japan, nine cases of zoonotic onchocercosis have been reported in Oita, Kyushu and in the western part of Honshu on the main island [5,10]. The causative agent of these infections was O. dewittei japonica [6,11] and the vector was identified as a blackfly Simulium bidentatum in Oita [12–14]. In this study, we present one clinical case found in a resident who lived in Iizuka City, Fukuoka Prefecture, Kyushu for some time.

A 24-year-old male developed a painful nodule on the middle finger of his right hand at the beginning of October 2011. He sought medical attention for the inflammation which had spread to the dorsal side of his right hand. The nodule was surgically removed from the vagina fibrosa tendinis of the finger at Beppu Medical Center, Beppu City, Oita Prefecture on January 10, 2012. The patient had lived in Iizuka City, Fukuoka Prefecture, in Kyushu, for 1 year and later moved to Beppu City in Oita Prefecture at the beginning of October 2011. Iizuka City is in a mountainous area inhabited by wild boars. He was a merchant who mainly imported vegetables and fruits from China and Southeast Asian countries and regularly visited fruit farmers in rural areas of northern Kyushu in connection with his business. Before he moved to Iizuka City, he had lived in Tochigi Prefecture on Honshu, the main island of Japan, and had visited the USA for a short period.

The mass excised (4 mm × 15 mm × 16 mm) was fixed in 20% buffered formalin and embedded in paraffin as a routine process in the laboratory of the hospital. Two histological sections were stained with hematoxylin and eosin (HE) and another section on a glass slide received Azan staining. After the surgery, he was examined for parasitic infections at the hospital and no signs were found. Based on histopathological examination, the mass consisted of a foreign-body granuloma and equipped with external transverse...
ridges which were salient and triangular (height, 10–20 μm and width, 20–25 μm) (Fig. 1A). The distance between two adjacent ridges was 100–115 μm. There were no inner striae in the middle layer of the cuticle (arrowhead in Fig. 1A). On the slightly oblique transverse sections, the thick portion of the cuticle constituted the ridges (arrows in Fig. 1B). The worm was surrounded with macrophages, neutrophils, eosinophils, lymphocytes, and collagen fibers (* in Fig. 1A, identified by Azan staining).

Seven Onchocerca species (O. dewittei japonica, O. gutturosa, O. cervicalis, Onchocerca eberhardi, Onchocerca lienalis, Onchocerca skrjabini, and Onchocerca suzukii) have been identified in domestic and wild animals in Japan [4]. In addition, two unnamed species (Onchocerca sp. type A from cattle and Onchocerca sp. from wild boar) were distinguished from other congeners by molecular analysis [13,14]. The distance between adjacent ridges of the causative agent was slightly shorter than that of O. dewittei japonica. The triangular ridges of this filarioid were identical to those of O. dewittei japonica. The ridges were larger than those of four other species (O. cervicalis, O. eberhardi, O. lienalis, and O. skrjabini) and different in shape from the rounded ones of O. gutturosa [6]. Females of O. suzukii lack transverse ridges and inner striae [5]. The causative agent was identical to O. dewittei japonica without inner striae, but differentiated from other congeners with inner striae. Therefore, we diagnosed the causative agent as a female O. dewittei japonica. The current case was the tenth recorded infection of O. dewittei japonica in Japan (Fig. 1C).

For molecular analysis, 30 worm tissues scraped from four unstained sections (10 μm thick) were used. The DNA extraction, polymerase chain reaction (PCR) amplification, and analysis of nucleotide sequences are described elsewhere [5,11]. The analysis was performed based on 110 bp of the mitochondrial cytochrome c oxidase subunit 1 (cox1) gene using MEGA 5.05 [15]. The phylogenetic tree was constructed based on GenBank sequences of cox1 of Onchocerca spp. using neighbor-joining (NJ) method (the accession number of this worm is LC051628), and the bootstrap probabilities were estimated. Two filarioids (Loxodontofilaria caprini and Cercopithifilaria longa) were used as outgroups.

Nucleotide sequences of the causative agent were identical to those of O. dewittei japonica, but different from those of six other congeners with the range being between 6.4% and 9.1%. Ferri et al. [16] suggested that the genetic distance of cox1 between two different species was greater than a threshold value of 4.8%. In the phylogenetic tree, the causative agent of this study and O. dewittei japonica had a high bootstrap value, which separated them from other congeners (Fig. 2). Therefore, we confirmed the causative agent as O. dewittei japonica.

Among the nine cases of zoonotic onchocercosis reported in Japan, the ages of patients ranged from 52 to 78, except for one case of a 2-year-old child. Eight female patients were recorded and one male [6]. In contrast, the current case of an infection in a male in his 20s was unusual. Many clinical cases of zoonotic onchocercosis indicated sporadic distribution worldwide. However, two infections of O. lupi were adenitically found in two patients, one lived in Istanbul while the other lived in both Istanbul and Edirne, Turkey [2,3]. Five previous cases were found in Oita Prefecture, which is 93 km × 114 km in size. The current infection was found in a resident of Iizuka City, Fukuoka Prefecture, which is the neighboring Prefecture of Oita Prefecture. In cases of infection of humans with Onchocerca volvulus, the time
between infection with the larvae and their growth to the adult stage is 10–20 months [17]. Therefore, we assumed that the patient was infected with O. dewittei japonica via blackfly while he was living in Iizuka City in Kyushu. These infections indicated that occurrences of zoonotic onchocercosis were concentrated in close range of northeast Kyushu. Similarly, three infections of the onchocercosis in Hiroshima Prefecture were found in a restricted spatial area (Fig. 1C) [18]. Global warming, unused rice fields, and decreasing numbers of hunters provide a favorable habitat for wild boars in Japan [5]. According to Otranto et al. [19], the high prevalence of *Thelazia callipaeda* or *Dirofilaria immitis* in dogs should be treated as an alert for risk of infection in the human population. The prevalence of *O. dewittei japonica* in wild boars was 89% in Oita [6]. *Simulium bidentatum* is the predominant blackfly species, being both zoophilic and anthropophilic, in Kyushu [4]. Therefore, this study suggests that a high incidence of transmission of *O. dewittei japonica* from wild boars to humans via blackflies has occurred in northeast Kyushu.

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References


