

## A Case Study of Human Infection with *Cysticercus cellulosae* (Cestoda: Taeniidae) Found in Okayama Prefecture, Japan

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**ABSTRACT.** An imported case of human cysticercosis found in Okayama Prefecture, Japan is reported. The patient (F.G.) is a 39-year-old male living in Okayama City who has immigrated into Japan as a Japanese orphan from Northern China early in March, 1993. On mid-April 1993, the patient noticed the presence of a small swelling on the skin surface of his right lower chest. He then visited Department of Dermatology, Okayama City Hospital on July 8th, 1993. Receiving an extirpation of the swelling a nodule with fibrous capsule was found in subcutaneous tissue. On histological examination, a single tapeworm larva having a protoscolex and numerous calcareous corpuscles was found in the capsulated nodule. The larval tapeworm found was identified as *Cysticercus cellulosae*, the pork tapeworm, *Taenia solium* Linnaeus, 1758 based on morphological characteristics of the rostellum armed with chitinous hooklets and cup-like muscular suckers on the protoscolex.

To our best knowledge, present report is the first case of human cutaneous cysticercosis in Okayama Prefecture, Japan.

**Key words:** imported human cysticercosis — *Cysticercus cellulosae* — *Taenia solium* — Cestoda — Okayama Prefecture

Human cysticercosis is known to cause by infection with larval form (*Cysticercus cellulosae*) of pork tapeworm, *Taenia solium* Linnaeus, 1758. Although man is an only definitive host for *T. solium* and usual intermediate hosts are pigs and wild boars, both adult worms and larval *C. cellulosae* can be found in the human body. *T. solium* is confined to be distributed tropical and subtropical countries where raw or insufficiently cooked pork meat is eaten. The patients infected with *T. solium* are recognized in many sites of the world, particularly in Eastern Europe, Central and South America, South Africa, Mongolia, China, Korea, and Russia.<sup>1,2)</sup>

Recently, human infection with *C. cellulosae* seems increasing in Japan, and about 400 cases have been reported in the literature.<sup>2,3)</sup> In Japan, the most reports of *C. cellulosae* infection are of cerebral cysticercosis, and cutaneous cysticercosis is exceedingly rare. Most patients of cutaneous cysticercosis

reported in Japan are exclusively limited to returned people from abroad, foreign entrants and immigrants from neighbouring countries, mainly China, Korea, Thailand or India. The authors wish to report a case of subcutaneous cysticercosis in immigrant from China who is now living in Okayama Prefecture together with bibliographical consideration.

### CASE REPORT

Patient : F.G., a 39-year-old male, an iron worker, living in Okayama City, Okayama Prefecture, Japan.

Main complaint : A small swelling on the skin surface of right lower chest.

Personal history : The patient has immigrated into Japan as a Japanese orphan from Northern China early in March, 1993. He lives alone in Okayama City, and he has occasionally eaten boiled fresh-water fishes such as *Carassius auratus* and *Cyprinus carpio* captured from Asahi River of Okayama Prefecture.

Present illness : On the middle of April 1993, the patient first noticed the presence of a small swelling on the skin surface of his right lower chest region. He then visited Department of Dermatology, Okayama City Hospital on July 8th, 1993. The cutaneous finding on the site was a freely movable indolent swelling of flesh to bright red color, 2 to 4 mm in diameter. The lesion was elevated 3 to 4 mm from the skin surface and diagnosed as Schwannoma. No other tumor was found in other regions of the body. Following day, the patient was submitted to the tumor operation, and a small nodule found in the subcutaneous tissue was excised.

TABLE 1. Laboratory data of the patient after the operation (July 15, 1993)

WBC	4,900/mm <sup>3</sup>	GOT	9 IU/L
St	4%	GPT	6 IU/L
Seg	56%	LDH	214 IU/L
Lym	32%	Amy	64 IU/L
Mon	3%	ALP	134 IU/L
Eo	5%	$\gamma$ -GPT	9 IU/L
RBC	$419 \times 10^4$ /mm <sup>3</sup>	LAP	44 IU/L
Hb	14.6 g/dl	ChE	0.62 pH
Ht	40.2%	BUN	21.0 mg/dl
Plt	$19.6 \times 10^4$ /mm <sup>3</sup>	UA	6.8 mg/dl
T-Bill	0.35 mg/dl	CRN	1.1 mg/dl
D-Bill	0.24 mg/dl	Na	146 mEq/L
TTT	1.9 Mu	K	4 mEq/L
ZTT	6.0 Ku	Cl	111 mEq/L
TG	85 mg/dl	Ca	4.6 mg/dl
		P	4 mEq/L

**Laboratory examination:** Prior to the operation no special sign was found in Wassermann reaction and anemia. Examination of the blood smears after the operation revealed that red blood cell count,  $419 \times 10^4/\text{mm}^3$ , hemoglobin level, 14.6 g/dl and white blood cell count,  $4900/\text{mm}^3$  with 4% stab form leucocytes, 56% segmented leucocytes, 32% lymphocytes and 5% eosinophils (Table 1). The fecal examination was negative for any ova and parasites.

**Past history:** The patient's family had been managing a fruit farm in China, and he had frequently been carrying feces of human and domestic animals as fertilizer of apple and pear trees. During the same time period, the patient had also worked on a breeding stock farm. He had experienced similar swelling on his neck region and received an operation several months before his arrival to Japan.

The present report is the first case of human infection with a *Cysticercus cellulosae* in Okayama Prefecture, Japan.

#### CYSTICERCUS CELLULOSAE

The nodule isolated from the subcutaneous tissue was ellipsoidal in shape, and the surface was yellowish-white in color. For histological examination the nodule was fixed in 10% formalin, sectioned at  $10 \mu\text{m}$  and stained with hematoxylin and eosin in a usual manner.

Microscopic observations showed that the circumference of the nodule was covered with a thick fibrous capsule (Fig 1). The circumferential capsule was

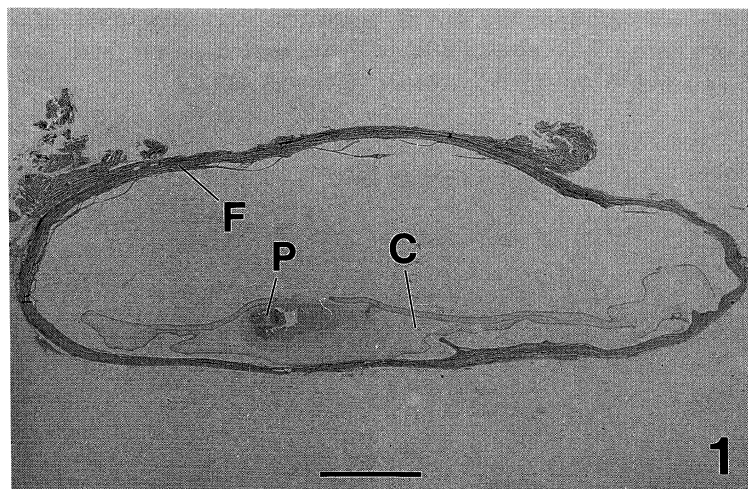


Fig 1. Cross section through a nodule, showing the fibrous wall and encapsulated *Cysticercus cellulosae* of *T. solium* (H&E, scale=3.0mm)  
C: *Cysticercus cellulosae*, F: fibrous wall, P: protoscolex of *C. cellulosae*

about 20 mm in largest dimensions and 6.5 mm in smallest ones. By histological examinations of the nodule the larval tapeworm with a single invaginated protoscolex approximately 0.6 mm in transverse diameter was found on one side of the capsule, but only after careful examination of subserial sections. It was found, furthermore, that one or two muscular suckers

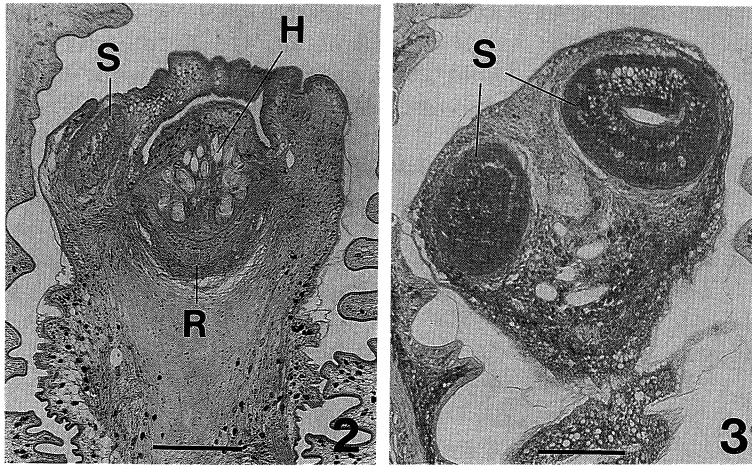


Fig 2, 3. Higher magnification of the portion of a single protoscolex, showing numerous hooklets on the rostellum and one of the 4 suckers (2), and 2 of the 4 suckers (3) (H&E, scale=20  $\mu$ m)

H: hooklets, R: rostellum, S: suckers

and numerous hooklets exist on the rostellum area of protoscolex (Fig 2, 3). Moreover, many small and oval-shaped calcareous corpuscles were found in the parenchyma just behind the protoscolex of the larval tapeworm (Fig 4). Fibrous wall surrounding the larval tapeworm was composed of connective tissue having numerous eosinophils, polynuclearleucocytes and macrophages, and measured about 0.3 to 0.5 mm in thickness (Fig 5).

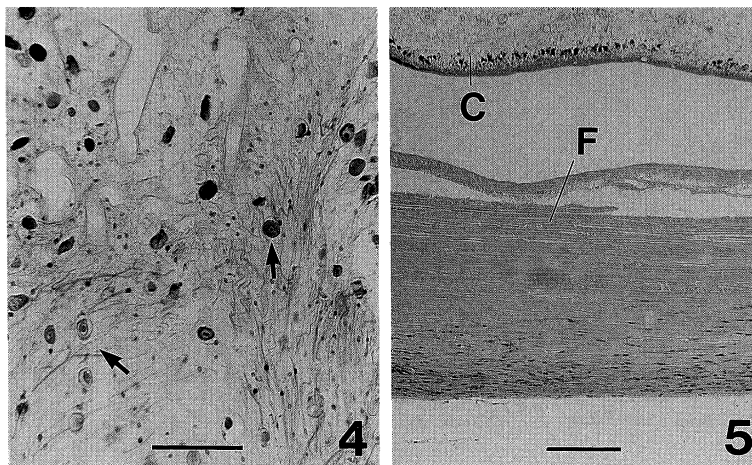


Fig 4. Higher magnification of parenchymal network just behind the protoscolex, showing small and oval shaped calcareous corpuscles (arrows) (H&E, scale=10  $\mu$ m)

Fig 5. Higher magnification of fibrous wall surrounding a *Cysticercus cellulosae* (H&E, scale=10  $\mu$ m)

C: *Cysticercus cellulosae*, F: fibrous wall

The larval tapeworm found in the nodule was identified as a *Cysticercus cellulosae hominis* based on morphological characteristics of protoscolex with suckers, hooklets, and calcareous corpuscles.

### DISCUSSION

It is well known that human is an only definitive natural host for pork tapeworm, and the larval stage of *T. solium*, *Cysticercus cellulosae* may also exist in the human body. The usual intermediate hosts of this tapeworm are pigs and wild boars. Sheep, camels, deer, dogs, monkeys, rats, and cats are less frequently infected, and human and other primates only occasionally (Brown, 1975).<sup>4)</sup> Among them pig is the most important infectious source for human infection. Individual of cysticercus in the body of the intermediate hosts most often invades into subcutaneous tissue, eye, brain, skeletal muscle, or heart ; but occasionally lung, liver, or kidney may be affected (Raimier and Wolf, 1978).<sup>5)</sup>

A main route of human infection with *T. solium* occurs when man ingests raw pork meat containing *C. cellulosae*. The larvae develop into adult worm in the host intestine. A route of human infection with *C. cellulosae*, on the other hand, occurs by accidental ingestion of the eggs excreted in infected human feces. The eggs hatch in small intestine and hexacanth larvae penetrate with its hooklets through the intestinal wall and enter the systemic circulation via hepatic portal vein and lymphatic vessels. The hexacanth larvae move through blood stream to the tissue where the hooklets are lost and the larvae develop into *C. cellulosae*. Growth of the larva in the human body is fairly rapid. In 3 weeks the developing larva is from 1 to 6 mm in length with visible protoscolex, and in 9 to 10 weeks it is fully developed infective bladder worm known as *C. cellulosae* (Belding, 1965).<sup>6)</sup> Brown (1975)<sup>4)</sup> has reported that man may incidentally acquire cysticercosis from the pork tapeworm eggs in three ways : (1) the ingestion of food or water contaminated by infected human feces, (2) oral transmission by the unclean hands of carriers of the adult worm, and (3) internal autoinfection by the regurgitation of eggs into the stomach by reverse peristalsis.

The present patient could not recall an exact date of accidental infection, but the authors are convinced that the infection occurred in China because he had resided in China almost all his life and had never left the country. In this patient, there were no sign of neurological symptoms or cerebral cysticercosis. Furthermore, it is thought that the infection with *C. cellulosae* was probably by oral ingestion of the eggs from infected human feces since no taeniid eggs were found from his own feces.

The first case of human infection with *C. cellulosae* in Japan was reported by Fukushima in 1908 of an Army soldier who returned from Manchuria.<sup>7)</sup> Thereafter related cases have extremely been increasing, although human cases of *C. cellulosae* infection have apparently decreased after 1955. Matsumura *et al* (1979)<sup>3)</sup> have summarized a total of 12 human cases of cutaneous cysticercosis in Japan from 1959 to 1977. According to them, the patients infected in Japan was only 3 but all other cases were assumed as foreign origins.

Imported cysticercosis can still be seen in Japan occasionally. A total of 14 cutaneous cysticercosis *cellulosae hominis* have been reported in Japan

between 1979 and 1994 as shown in Table 2. Twelve out of 14 patients (Table 2) are imported cases from neighbouring countries; 6 in Korea,<sup>11,14,16,18)</sup> 4 in China<sup>8,12,13,15)</sup> and one each in India<sup>11)</sup> and Thailand.<sup>17)</sup> As evident from Table 2, the patients of cysticercosis infected in Japan are only 2 cases.<sup>9,10)</sup> These patients had not been abroad, and route of infection of these patients is not definite. Taking into account the increasing phenomena of returned people from abroad and travelers from overseas, as well as imported pigs, continuous increase of the *C. cellulosae* infection is expected in Japan in the near future.

Therefore special attention should be given to this type of infection. The diagnosis of cutaneous tumor caused by *C. cellulosae* is extremely difficult to distinguish from other one. Hence, the authors emphasize necessity of careful histological examination for cutaneous nodule in addition to radiography, computerized tomography and immunological tests when cysticercosis cellulosae hominis is suspected.

TABLE 2. Cutaneous cysticercosis reported in Japan between 1979 and 1995

Cases	Patients			Lesion sites	Sites of infections	Authors(Year)
	ages	sexes	residences			
1	53	F	Kitakyusyu	Left lower thigh	China	Fukui <i>et al</i> (1979) <sup>8)</sup>
2	43	"	Saitama	Left thigh	Saitama*	Ishibashi <i>et al</i> (1980) <sup>9)</sup>
3	34	"	Osaka	Left lower thigh	Osaka*	Tanoue <i>et al</i> (1981) <sup>10)</sup>
4	31	M	Kyoto	Neck, Body	Korea	Yoshida <i>et al</i> (1982) <sup>11)</sup>
5	28	"	"	Neck	India	
6	38	F	"	Left forearm	Korea	
7	46	M	Saitama	Whole body	China	Hori <i>et al</i> (1982) <sup>12)</sup>
8	53	"	?	Left clavicule	China	Hiraiwa <i>et al</i> (1983) <sup>13)</sup>
9	44	"	Fukuoka	Arm, Thigh	Korea	Nakashima <i>et al</i> (1984) <sup>14)</sup>
10	62	F	?	Whole body	China	Hiraki <i>et al</i> (1987) <sup>15)</sup>
11	44	M	?	Both thigh	Korea	Nakai <i>et al</i> (1990) <sup>16)</sup>
12	29	F	?	Mandible	Thailand	Sato <i>et al</i> (1991) <sup>17)</sup>
13	48	"	?	Whole body	Korea	Kuboyara <i>et al</i> (1994) <sup>18)</sup>
14	43	F	?	Whole body	Korea	
15	39	M	Okayama	Right lower chest	China	Present authors

F=female, M=male, \*=the patient has not been abroad.

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