Epidemiol. Infect. (2014), **142**, 2654–2661. © Cambridge University Press 2014
doi:10.1017/S0950268814000144

Retrospective epidemiological analysis of sparganosis in mainland China from 1959 to 2012

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Received 14 May 2013; Final revision 16 December 2013; Accepted 15 January 2014; first published online 4 February 2014

SUMMARY

In this study, epidemiological factors of sparganosis cases reported in mainland China from 1959 to December 2012 were analysed. A total of 1061 valid cases were distributed throughout most of the provinces of mainland China, with most cases occurring in Southern and Eastern China. The average age of patients was 29 years (range 0–80 years). Modes of transmission to humans were via contact (54·6%), mainly by application of frog meat as a poultice, foodborne (33·8%), mainly through ingesting frogs or snakes, and waterborne (11·5%) through drinking raw water. The tissue/organs involved were subcutaneous/muscle (43·1%), eyes (31·0%), central nervous system (CNS) (17·9%), urogenital system (3·9%) and visceral organs (3·2%). Obvious differences existed in main risk factors for different areas. Close correlation was found between tissue/organs and risk factors. Main modes of transmission changed during the past decades, from contact (83·8% pre-1979) to foodborne (63·9% post-2000). The tissue/organs involved also changed at the same time. Cases involving eyes fell from 50·0% pre-1979 to 8·3% post-2000, and cases involving CNS increased from 0% pre-1979 to 47·8% post-2000. These results illustrate that China is one of the main epidemic countries of sparganosis in the world. Consumption of frog/snake meat was the main risk factor, although application of frog flesh as a poultice was the main risk factor before 2000. Sparganosis has become one of the neglected but important foodborne/waterborne parasitic diseases in mainland China.

Key words: Epidemiology, parasites, risk assessment, tapeworms.

INTRODUCTION

Sparganosis or plerocercoidosis is a parasitic disease caused by the plerocercoid, the third larval stage of diphyllobothroid tapeworms belonging to the genus *Spirometra*, most commonly *Spirometra erinaceieuropaei* (also called *S. erinacei* or *S. mansoni*) in Asia [1, 2] and *Spirometra mansonoides* in North America [3]. Adult *Spirometra* worms parasitize the intestines of dogs and cats, and rarely humans (spirometrosis). The parasites exploit two intermediate hosts during its life-cycle: copepods as the first intermediate hosts, where the procercoid larva develops, and varieties of vertebrates (amphibians, reptiles, birds, mammals) as the second intermediate hosts, where the plerocercoid larva develops. In nature, frogs mainly act as second intermediate hosts, and frog-eating predators, such as snakes and birds, are paratenic hosts [3]. The parasite is transmitted to humans in three different ways: waterborne (drinking raw water contaminated with

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Fig. 1. Geographical distribution of sparganosis in mainland China.

copepods carrying procercoids), foodborne (ingesting raw or undercooked meat of the second intermediate hosts or paratenic hosts carrying plerocercoids) and contact (mainly by the application of frog flesh as a poultice in traditional medicine in several countries in Asia) [4, 5].

Plerocercoids can invade subcutaneous tissue/muscle, eyes, brain, urinary tract, pleura, spinal canal and other tissue/organs of the human body, leading to various clinical symptoms according to the location of the parasites within the host [6]. Misdiagnoses of sparganosis are rather common and most cases are diagnosed following surgical removal of the worm, especially in cases of infections of the central nervous system (CNS) and visceral organs [7, 8]. Although immunodiagnostic and CT/MRI methods have been developed, identification of risk factors has significant value for pre-operative diagnosis. Surgical removal of the worms is the best treatment of this disease, in contrast, medication (praziquantel, mebendazole, etc.) has limited success. Prevention strategies should focus on food and water sanitation and against using raw frog or snake flesh as poultices [9].

Table 1. *Age distribution of 504 cases in mainland China*

Age range (yr)	Cases (*n*)	Ratio (%)
0–10	81	16·1
11–20	94	18·7
21–30	110	21·8
31–40	95	18·8
41–50	55	10·9
51–60	44	8·7
>60	25	5·0
Total	504	100·0

The first human case of sparganosis was reported by Manson in 1882 in China [2], after that the disease

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Table 2. *Tissues/organs involved in 1061 cases in mainland China*

Tissues/organs	Cases (*n*)	Ratio (%)
Subcutaneous/muscles		43·1
Maxillofacial region	173	16·3
Cervical region	8	0·8
Thoracic region	66	6·2
Breast	37	
Chest wall	20	
Axillary region	5	
Thorax-abdominal region	4	
Upper limb	13	1·2
Waist/back	10	0·9
Abdomen	70	6·6
Inguen	16	1·5
Lower limbs	76	7·2
No detailed location	25	2·4
Eyes	329	31·0
Central nervous system	190	17·9
Brain	182	
Spinal cord	4	
Vertebral canal	4	
Genitourinary system	41	3·9
Epididymis	2	
Testis	2	
Spermatic cord	4	
Urinary canal	1	
Labia vulvae	9	
Penis	8	
Scrotum	15	
Visceral organs	34	3·2
Abdominal cavity, chest cavity, liver, lung, kidney, etc., most involved multi-organs		
No detailed information recorded	10	0·9
Total	1061	100·0

was found widely distributed in many tropical and subtropical countries, especially in East and Southeast Asia. China is one of the main countries to suffer from sparganosis. In the present study, the epidemiology of sparganosis in mainland China from 1959–2012 was analysed.

MATERIALS AND METHODS

The literature relating to sparganosis in China (not including Taiwan, Hong Kong, Macao), from 1959 to December 2012 was collected from online databases including China National Knowledge Infrastructure (CNKI, http://www.cnki.net/), Chinese Medical Current Contents (CMCC), Wanfangdata (http://g.wanfangdata.com.cn/) and other Chinese periodicals using the key words 'sparganosis' and 'plerocercoidosis'. All cases of sparganosis were collected, epidemiological factors such as reporting time, geographical distribution, age and gender of patients, urban/rural distribution, possible risk factors, and tissue/organs involved in each case were extracted to form a table using Microsoft Office Excel 2003; data were analysed by the ratio of percentage (all cases were anonymized for analysis).

RESULTS

Valid cases

Over the period studied, 1114 cases were collected from 411 reports published in 215 periodicals. A total of 52 cases that were repeated in more than one report were excluded. One case that was confirmed as proliferative sparganosis was not included in this

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study [10]. A total of 1061 valid cases accounting for 95·2% of all cases was used in the analysis. Of the valid cases 92·4% (980/1061) were confirmed by pathological observation or direct observation of parasites post-operation, and the remainder (7·6%, 81/1061) were confirmed by immunological diagnostic methods (skin test, ELISA) combined with the patients' history.

Geographical distribution

Sparganosis was distributed throughout most provinces apart from Inner Mongolia, Shanxi, Tianjin, Gansu, Shaanxi and Tibet (Fig. 1). Nearly 60% of the cases were distributed in the southeast coastal areas (Southern China 31·2%, Eastern China 25·8%). The top five provinces with the highest case load were Guangdong (171), Hunan (112), Hainan (104), Jilin (94), Fujian and Sichuan (89); the top five provinces with the highest incidence were Hainan (1·2), Jilin (0·3), Shanghai (0·2), Fujian (0·2) and Hunan (0·2) according to incidence (1/100 000) calculated from 2012 population data of every province (data not shown).

Sex, age and urban/rural residence of the patients

No obvious difference in sex was found (424 male, 465 female) in the 889 cases with gender recorded in all valid cases. Patients' age ranged from newborn to 80 years in 504 cases with definite age records, the average age was 29 years and most patients were in the 0–50 years age group, accounting for 86·3% (Table 1). Of the 360 cases with occupational records, most patients (74·7%) were from rural areas, with the remainder were from urban areas.

Tissue/organs and risk factors

For the 1061 valid cases, the most commonly involved tissue/organs were: subcutaneous/muscles (43·1%), followed by eyes (31·0%), CNS (17·9%), genitourinary system (3·9%), visceral organs (3·2%) and no detailed information recorded (0·9%). Details of the tissue/organs involved are listed in Table 2. In 884 cases with risk factors recorded, the modes of transmission were through contact (54·6%), foodborne (33·8%) and waterborne (11·5%). Details of the risk factors are listed in Table 3.

Table 3. *Mode of transmission and risk factors of 884 cases in mainland China*

Transmission mode/risk factors	Cases (*n*)	Ratio (%)
Contact	483	54·6
Applying frog meat as a poultice	441	
Applying chicken gall as a poultice	1	
Applying herbal medicine as a poultice	9	
Contact with frogs or tadpoles	27	
Contact with contaminated water	5	
Foodborne	299	33·8
Consuming raw meat of frog, snake	258	
Swallowing tadpoles	32	
Swallowing snake gall	6	
Swallowing chicken gall	1	
Eating raw pork	2	
Waterborne		
Drinking raw water	102	11·5
Total	884	100·0

Correlation of the mode of transmission with geographical distribution and tissue/organs

Modes of transmission for the different regions are listed in Table 4. In Northeastern China, Northern China and Central China, the main mode of transmission was foodborne (41·6%, 66·7% and 62·4%, respectively), with most patients having a history of ingesting frog or snake meat. Compared to that, in Eastern China, Southern China and Southwestern China, the main mode of transmission was through contact, especially the application of frog meat as a poultice (53·1%, 74·6% and 67·5%, respectively).

Mode of transmission for cases with the tissue/organs involved is shown in Table 5. For cases of maxillofacial, eye and upper limbs, the main mode of transmission was through contact, mainly the application of frog meat as a poultice (90·4%, 90·1% and 76·9%, respectively). For cases concerning other areas of body, except breast, the main mode of transmission was foodborne, mostly the consumption of raw frog or snake meat.

Tissue/organs and mode of transmission in different periods

The main tissue/organs involved changed markedly over the period studied. Cases involving eyes decreased from 50·0% pre-1979 to 8·3% post-2000. Cases involving subcutaneous/muscles also exhibited

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Table 4. *Correlation of risk factors with geographical distribution*

	Contact		Foodborne		Waterborne		Total	
Area	No.	(%)	No.	(%)	No.	(%)	No.	(%)
Northeastern China	24	(23·8)	42	(41·6)	35	(34·7)	101	(100)
Northern China	2	(22·2)	6	(66·7)	1	(11·1)	9	(100)
Eastern China	111	(53·1)	77	(36·8)	21	(10·0)	209	(100)
Southern China	223	(74·6)	52	(17·4)	24	(8·0)	299	(100)
Central China	44	(31·2)	88	(62·4)	9	(6·4)	141	(100)
Northwestern China	2	(20·0)	4	(40·0)	4	(40·0)	10	(100)
Southwestern China	77	(67·50)	31	(26·3)	7	(6·1)	115	(100)
Total	483	(54·6)	300	(33·9)	101	(11·4)	884	(100)

Table 5. *Relationship of mode of transmission with tissue/organs*

	Contact		Foodborne		Waterborne		Total	
Tissue/organ	No.	(%)	No.	(%)	No.	(%)	No.	(%)
Subcutaneous/muscles								
Maxillofacial	150	(90·4)	12	(7·2)	4	(2·4)	166	(100)
Neck	3	(50·0)	3	(50·0)	0	(0·0)	6	(100)
Chest	1	(5·0)	13	(65·0)	6	(30·0)	20	(100)
Breast	7	(25·0)	10	(35·7)	11	(39·3)	28	(100)
Upper limb	10	(76·9)	0	(0·0)	3	(23·1)	13	(100)
Waist/back	1	(14·3)	6	(85·7)	0	(0·0)	7	(100)
Abdomen	5	(8·5)	37	(62·7)	17	(28·8)	59	(100)
Inguen	1	(6·3)	9	(61·5)	6	(30·8)	16	(100)
Lower limb	11	(15·3)	39	(62·9)	12	(19·4)	62	(100)
Others	2	(9·1)	20	(90·9)	0	(0·0)	22	(100)
Eyes	274	(90·1)	19	(6·3)	11	(3·6)	304	100·0
Central nervous system	5	(4·5)	77	(70·0)	28	(25·5)	110	100·0
Genitourinary system	8	(23·5)	22	(64·7)	4	(11·8)	34	100·0
Visceral organs	2	(6·9)	24	(82·8)	3	(10·3)	29	100·0
No detailed information recorded	2	(25·0)	6	(75·0)	0	(0·0)	8	100·0
Total	482	(54·5)	297	(33·6)	105	(11·9)	884	100·0

a decrease; while, cases involving CNS and visceral organs increased, especially for CNS, from no case reported pre-1979 to 47·8% post-2000. Most CNS cases (85·3%, 162/190) were reported during this period (Table 6). The main modes of transmission in different periods also changed, cases caused through contact decreased gradually, from 83·8% pre-1979 to 18·1% post-2000; while foodborne cases increased, from 7·3% pre-1979 to 63·9% post-2000; waterborne cases also increased (Table 7).

DISCUSSION

There have been more than 1700 global records of sparganosis to date (mainland China, ≈1000 cases [6]; Japan, 470 cases [11]; South Korea, ≈200 cases [12]; Thailand, 52 cases [13]). Sparganosis caused by *S. erinaceieuropaei* is dominant in South and East Asia [14]. A total of 1061 cases analysed in the present study account for about 60·0% of the global records, and around 60·0% of the cases distributed along the southeast coast of mainland China. Teenagers and adults from rural areas are the main groups experiencing infection, and no obvious sex difference was observed.

The primary mode of sparganosis transmission in mainland China during the survey period was via contact (54·6%), with most cases infected through the use of frog flesh as a poultice for self-treatment. This contact transmission was mainly distributed in

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Table 6. *Tissue/organs involved in different periods*

	S/M		E		CNS		GS		VO		Total	
Period	No.	(%)	No.	(%)	No.	(%)	No.	(%)	No.	(%)	No.	(%)
Up to 1979	92	(46·5)	99	(50·0)	0	(0·0)	5	(2·5)	2	(1·0)	198	(100)
1980–1999	256	(49·8)	202	(39·3)	28	(5·4)	25	(4·9)	3	(0·6)	514	(100)
2000 onwards	110	(32·4)	28	(8·3)	162	(47·8)	10	(2·9)	29	(8·6)	339	(100)
Total	458	(43·6)	329	(31·3)	190	(18·1)	40	(3·8)	34	(3·2)	1051	(100)

S/M, Subcutaneous/muscles; E, eyes; CNS, central nervous system; GS, genitourinary system; VO, visceral organs; UN, no detailed information recorded.

Table 7. *Mode of transmission in different periods*

	Contact		Foodborne		Waterborne		Total	
Period	No.	(%)	No.	(%)	No.	(%)	No.	(%)
Up to 1979	150	(83·8)	13	(7·3)	16	(8·9)	179	(100)
1980–1999	288	(63·2)	127	(27·9)	41	(9·0)	456	(100)
2000 onwards	45	(18·1)	159	(63·9)	45	(18·1)	249	(100)
Total	483	(54·6)	299	(33·8)	102	(11·5)	884	(100)

Southern, Southwestern and Eastern China. This mode of transmission is quite different from other countries. In South Korea and Japan, the main mode was foodborne. Consuming the flesh of snake or fowl has been considered to have nutritional and medicinal effects for ill and weak people in these countries [4]. In Thailand, most patients were infected by drinking raw water contaminated with infected copepods, although there were a few cases caused by application of frog or snake flesh as a poultice [15]. Risk factors of mode of transmission in different areas varied in China. For example, the most common risk factor was using frog flesh as a poultice to cure inflammation of the eyes and body surface in Southern and Eastern China [16–23]. In Sichuan province, most patients became infected through biting and retaining frog flesh in the mouth as a cure for toothache [24]. There were nine cases in Southern China that had a history of using herbs, i.e. heartleaf *Houttuynia* herb. This herb grows along water-edges and has detoxification and antipyretic properties in traditional Chinese medicine, and is used as a poultice [18, 25]. It was speculated that the herbs were contaminated by procercoids or copepods in water. Most cases infected by contact occurred in rural areas before 2000 due to poor medical conditions and local customs.

The second mode of transmission was foodborne, mainly caused by ingesting undercooked meat of the second intermediate host such as frog, snake and other animals [26]. There were a few cases infected by special risk factor. For example, most cases (26/34) reported in Henan province were said to be caused by the unusual practice of swallowing live tadpoles to 'relieve inflammation or internal heat', and all these cases were confirmed by ELISA [27]. In traditional Chinese medicine, the gallbladder of the snake was believed to have detoxification and antipyretic properties. Ingestion of the gallbladder or drinking bile with wine was a popular practice with people consuming snake. There were six cases infected in this way and one case infected by swallowing the gallbladder of chicken [28–31].

The third mode of transmission was waterborne. In rural areas, many farmers were accustomed to routinely drinking untreated water. If the water was contaminated with copepods and/or procercoids, infection might occur.

In 177 cases with no obvious/unknown medical histories, there was one case in a newborn child. Shortly after the child was born, an uplift wound was found at the left ear hairline, from which a plerocercoid of length of 475 mm was removed. It has been established that this disease can also be caused by vertical transmission [32].

Most cases of sparganosis located in the maxillofacial region, eyes and upper limbs, were caused by

https://doi.org/10.1017/S0950268814000144 Published online by Cambridge University Press

contact, especially the use of frog flesh as a poultice, accounting for 90·4% (150/166), 90·1% (274/304) and 76·9% (10/13) of cases, respectively. However, for cases of other parts of the body, the first mode of transmission was foodborne followed by waterborne. This implies that there is a close correlation between disease location and mode of transmission.

In past decades great changes have occurred in China regarding the economy, medical and health conditions, popularity of heath knowledge and improved living standards. Customs such as using frog meat as a poultice or swallowing tadpoles for self-treatment were rare in epidemic areas, while the consumption of frogs, snakes, and other wild animals were popular in some areas of China. Therefore, cases caused by contact decreased markedly (from 83·8% pre-1979 to 18·1% post-2000), while foodborne cases increased (from 7·3% to 63·9% in same period), and cases caused by drinking contaminated water also increased. Sparganosis has become one of the important food-/waterborne parasitic diseases in China [33].

Most cases in the present study were confirmed by post-operative biopsy. Although there were immunodiagnostic and CT/MRI methods for the auxiliary diagnosis [34–36], they are not popular in most hospitals in China, especially before 2000. Besides the cases discussed in this study, it is believed that there were more cases not reported due to misdiagnosis or other reasons. Before the 1980s, due to the absence of effective diagnostic methods, there was no report of CNS sparganosis in China. The first domestic case of CNS was reported in 1982 [37]. After the 1980s, CT/MRI technology was introduced in some hospitals in China, which greatly contributed to surgical therapy for CNS occupied lesions and more sparganosis CNS cases were confirmed by pathological observation or direct observation of parasites post-operatively, but misdiagnosis remained at a high level pre-operatively. In the past decades, CNS sparganosis is the most common of cases reported, accounting for 47·8% of all sparganosis cases [38–40].

In conclusion, China is the main epidemic country of sparganosis in the world, the disease is distributed throughout most provinces especially in Southern and Eastern China. The main mode of transmission is via contact, followed by foodborne and waterborne transmission. There are great differences in the main risk factors of sparganosis in different areas due to the customs and eating habits. The tissue/organs involved have a close relationship with the risk factors. The main risk factors have changed over the past decades, from using raw frog flesh as a poultice to consuming frog and snake meat. The tissue/organs involved also changed at the same time, cases of oral/maxillofacial, and eyes decreased and cases of other parts of the body, especially CNS, increased. Now, sparganosis has become one of the neglected but important foodborne/waterborne parasitic diseases in China. In future, not consuming the raw flesh of snakes, frogs, fowl or other animals and not drinking raw water will be the main strategies to prevent sparganosis. Early and effective diagnostic methods and new medications should be developed.

ACKNOWLEDGEMENTS

This work was supported by the National Nature Science Foundation of China (grant no.81260254).

DECLARATION OF INTEREST

None.

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