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The first case of human sparganosis in Poland and a review of the cases in Europe

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1. Introduction

Sparganosis is a zoonotic disease caused by infection with a larval form of *Spirometra* species tapeworms. The first intermediate host is freshwater cyclops (copepods) and the second intermediate hosts are mainly amphibians. An array of frog-eating snakes, birds and mammals can be a paratenic host harbouring plerocercoids, an infective stage of tapeworm, also known as sparganum. An adult tapeworm inhabits the intestinal tract of household and wild canidae and/or felids. Human infection can occur in two ways, drinking water contaminated with copepods harbouring procercoid larvae, or consuming raw or undercooked meat of the second intermediate or paratenic hosts, mostly frogs. Snakes or chicken [1–3]. In addition, infection can also happen via percutaneous route, which has been documented in southern China and Sotheast Asia, where frogs and snakes have been used anti-inflammatory poultice [4]. In humans, the *Spirometra* sp. larvae usually resides in the subcutaneous tissues and does not develop further into its adult form [1–4]. Although it is rare, *Spirometra* larvae migrate in to vital organs such as eyes or central nervous system to cause deleterious lesions [1–4]. The parasite can survive in the human tissue up to 20 years [1–3]. Human sparaganosis is a common zoonotic parasitosis in Asian countries; over 1000 cases were recorded from China [3], over 500 cases from Japan [5], over 400 cases from Korea [6] and nearly 100 cases from Thailand [4]. Sporadic cases have been reported from many Asian countries [1–3]. Human sparganosis cases have also been reported from North [7] and South America [8] and some countries of Africa [9]. In Europe, however, human sparganosis cases are extremely rare (see review in the Discussion of this article) and is quite an unfamiliar disease. Here we report the first case of human sparganosis in Poland with the comprehensive review of the cases in the Europe.

2. Case description

A 60 year-old caucasian woman living in Hajnówka, north-eastern Poland, came to the Oncologic Surgery Outpatient Clinic in Hajnówka in March 2016 with two detectable subdermal noduler lesions lying close to each other. According to the anamnesis, the patient has never left Poland to go abroad, and she moved around maximum 200 km radius from her dwelling-place through her life. Patient did not report any other diseases. She said that she had eaten venison several times. On physical examination the nodular lesions on the right lumbar abdominal region was confirmed. The patient stated that she had a tick-bite in November 2015. The attending doctor ordered fine needle aspiration (FNA) and histopathological examination. The ultrasonography (US) showed an image of a concentric heterogeneous echogenicity distinct from the surrounding fatty tissue (Fig. 1). The material obtained from the biopsy was light grey, watery and turbid. The microscopic examination of the smear showed the presence of inflamed tissue fragments, multinucleated giant cells and gelatinous shreds surrounded with eosinophils. Due to the unique nature of the US image and lack of the definitive diagnosis after FNA, the physicians decided surgical extirpation of the whole nodular lesion and to conduct histopathological examination for the surgical specimen.

2.1. Macroscopic assessment of the surgical materials

Surgically removed materials in two dishes was handed for histopathological examination:

Dish 1 – A piece of skin tissue 2 × 2 × 1.5 cm covered with epidermis on one side 2 × 1 cm and the second piece of 2 × 2 × 1 cm with epidermis of 2 × 0.8 cm.
Dish 2 – a parasite or a parasite's larva of about 17 cm in length, 0.5 cm wide, flattened, hard, cream-white in colour resembling a 'wrinkled ribbon' by the naked eye observation (Fig. 2). The front end was wider than the rest of the body and had two longitudinal grooves (bothria) (Figs. 2,3).

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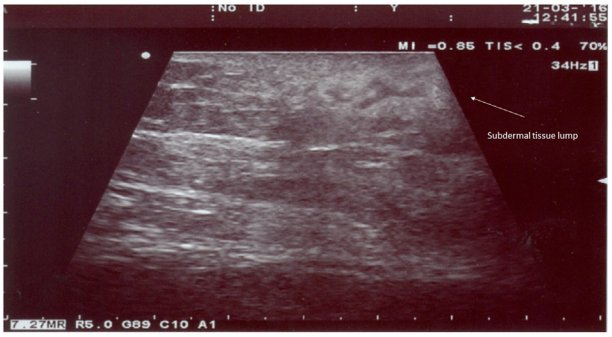

Fig. 1. An ultrasonographic image of subcutaneous lesion of the patient.

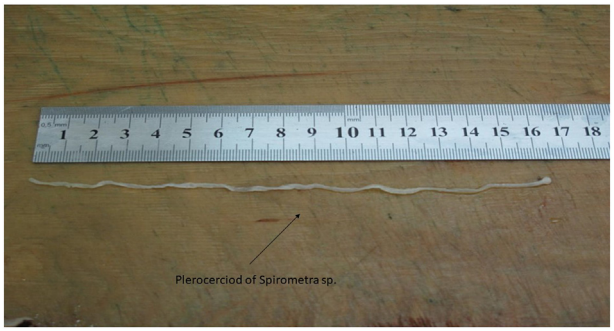

Fig. 2. Morphological appearances of *Spirometra* sp. plerocercoid isolated from the skin lesion of the patient.

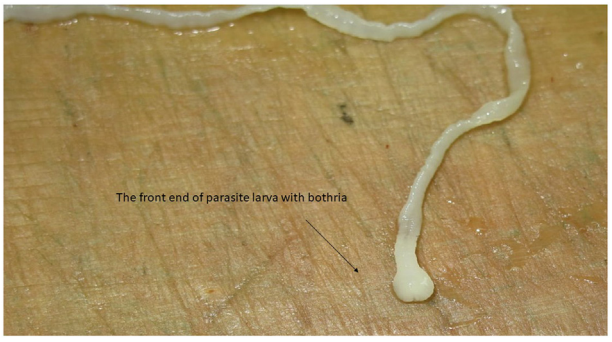

Fig. 3. Close-up view of the anterior portion of the worm.

2.2. Microscopic assessment of tissues

The skin tissue covering the nodular lesions has no remarkable pathological changes. Chronic, intensive inflammatory infiltrates with the areas of fatty tissue necrosis were present in the subdermal tissue. In the vicinity of the cystic area where the parasite was, there was a granulomatous lesion with multinucleate giant cells.

2.3. Microscopic appearances of the parasite

Sections of the worm were stained with hematoxylin and eosin. The body of the parasite showed typical tegument structure of tapeworms. The parasite has no segment, no intestine, and had a homogeneous internal structure. With higher magnification, eosinophilic tegument with the nucleus layer and an internal watery stroma, which contained thin myofibres and round cells, were found (Fig. 4). Taking all these macroscopic and microscopic appearances, the parasite was identified as a plerocercoid form of Spirometra sp. plerocercoid.

Because the tapeworm larva was placed in formalin immediately after surgical removal, further genetic identification of the species was difficult to perform.

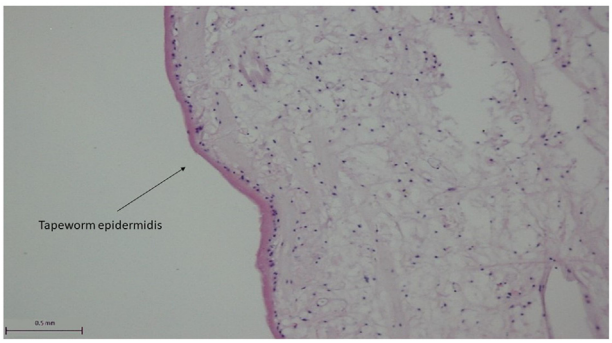

Fig. 4. Cross-section of the *Spirometra* sp. plerocercoid (H-E staining) An arrow indicate the tegument.

3. Discussion

As briefly reviewed in Introduction, human sparganosis is common in East and Southeast Asia, especially in China, Japan, Korea and Thailand [3–6], and sporadically in North and South America [7,8] and some places of Africa [9]. However, human sparganosis has rarely been documented in Europe. The first indigenous human sparganosis case in Europe was recorded from Italy in 1953 [10], and Pampiglion et al. (2003) added 4 more cases including their own case from Italy [11–14] and 2 cases from France [15,16]. Subsequently three more cases, one case each from Czech [17], Germany [18] and Italy [19], were added. The recent case from Germany [18] was supposed to be infected with *Sparganum proliferum* during his trip to Bolivia, Brazil and Paraguay. Thus, our case reported here is the eleventh case in Europe.

In addition to those 10 cases, a few cases of sparganosis have been reported among immigrants living in Europ from endemic countries; a case of cutaneous sparganosis in Vietnamese in Czhech [20] and a case Japanese woman in Germany [21]. A case of ocular sparganosis was found in a Sri Lankan immigrant in France [22] and four cases of cerebral sparganosis, a Malawian in Germany [23], a Bangladeshi in Germany [24], a Chinese in eastern England [25] and a Bolivian in Spain [26]. All those indigenous and immigrants cases of human sparganosis in Europe are summarized in Table 1.

As the main and popular source of infections of human sparganosis,

Table 1
Human sparganosis cases in Europe.

No.	Year	Country	Gender	Age	Lesion	Reference
Indigenous cases						
1	1953	Italy	F	40	Forarm skin	10
2	1953	Italy	M	28	Groin skin	11
3	1964	Italy	M	30	Subclavian skin	12
4	1976	Italy	M	28	Brain	13
5	1997	France	M	21	Chest wall skin	15
6	1999	France	M	14	Ocular	16
7	2002	Italy	M	50	Thigh skin	14
8	2006	Czech	M	14	Ocular	17
9	2014	Germany	M	61	Chest wall skin[a]	18
10	2015	Italy	M	56	l-Elbow skin	19
11	2018	Poland				Present case
Immigrant cases						
1	1995	Czech (VNM)	M	29	r-Pectoral skin	20
2	2003	France (SriLanka)	M	17	Ocular	21
3	2007	Germany (Malawi)	M	25	Brain	22
4	2013	Germany (Japan)	F	68	abdominal skin	23
5	2013	Swiss (Bangladesh)	M	39	Brain	24
6	2014	UK (China)	M	50	Brain	25
7	2015	Spain (Bolivia)	M	29	Brain	26

[a] Sparganum proliferum infection during travel overseas.

consumption of raw or undercooked meat of amphibians, reptiles and/or of intermediate birds and mammals are well-known [1–3]. Alternatively, drinking of natural water contaminated with cyclops harbouring procercoid is possible when the patients have no clear history of consumption of raw meat of intermediate/paratenic hosts [1–3]. In the present study, the patient has never been to abroad and moved around 200 km from her dwelling-place all her life, so that she has no chance of infection in overseas. Conversely, *Spirometra* infection was found in the wild animals in the forests close to her residential area [27–29]. Even one new species, *Spirometra janicki* was reported from carnivorous mammals in Poland [30], its validity needs further study. Since the patient reported here has been living in such an environment, she might be infected by drinking outdoor water containing copepods harbouring *Spirometra* procercoids. Alternatively, since she had a history of consuming venison several times, it would be a possible source of infection. Since Spirometra plerocercoids were found in the muscles of wild boar in Poland [28], possibility of wild deer as a paratenic host for *Spirometra* should be explored urgently. Related to this, venison was proven to be a source of lung fluke infection in Japan [31]. More strict inspection and legal regulation is necessary for the wild meat consumption. Also, diagnostic tools to detect *Spirometra* sp. infections should be introduced in the hospitals and diagnostic laboratories in this country.

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Declaration of interest

None.

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References

[1] I. Miyazaki, Helminthic Zoonoses, International Medical Foundation of Japan, Tokyo, 1991.
[2] M.A. Marty, R.C. Neafie, Diphyllobothriasis and sparganosis, in: W.M. Meyers (Ed.), Pathology of Infectious Diseases. Vol 1 Helminthasis, Armed Forces Institute of Pathology, American Registry of Pathology, Washington DC, 2000.
[3] Q. Liu, M.W. Li, Z.D. Wang, G.H. Zhao, X.Q. Zhu, Human sparganosis, a neglected food borne zoonosis, Lancet Infect. Dis. 15 (2015) 1226–1235.
[4] M. Anantaphruti, Y. Nawa, Y. Vanvanitchai, Human sparganosis in Thailand: an overview, Acta Trop. 118 (2011) 171–176.
[5] N. Kagei, Food-borne parasitic diseases: Sparganosis mansoni, SRL Ho-kan 20 (3) (2000) 115–121 (in Japanese).
[6] J.G. Kim, C.S. Ahn, W.M. Sohn, Y. Nawa, Y. Kong, Human sparganosis in Korea, J. Korean Med. Sci. 33 (44) (2018 Sep 21) e273, https://doi.org/10.3346/jkms.2018.33 (e273. eCollection 2018 Oct 29).
[7] W.G. Strauss, J.H. Manwaring, Sparganosis: a new case in the United States and review of the literature, Int. J. Dermatol. 3 (2) (1964) 73–78, https://doi.org/10.1111/j.1365-4362.1964.tb06023.x.
[8] P.A. Rolon, Report of a probable 7th case in South America and review of the subject, Bull. Soc. Pathol. Exot. 69 (4) (1976) 351–359 Human sparganosis.
[9] M.L. Eberhard, E.A. Thiele, G.E. Yembo, M.S. Yibi, V.A. Cama, E. Ruiz-Tiben, Thirty-seven human cases of sparganosis from Ethiopia and South Sudan caused by Spirometra spp, Am. J. Trop. Med. Hyg. 93 (2) (2015) 350–355.
[10] D. Pujatti, Un caso di sparganosi umana, Riv. Parassitol. 14 (1953) 213–217 (in Italian).
[11] G. Rossi, M. Genesi, Pseudotumori parassitari, Minerva Med. 44 (1953) 1517–1525 (in Italian).
[12] L. Bianchi. Sparganosi umana. Osservazione sopravun caso autoctono in Alta Italia. Proc First Intern Congr Parasitol, Rome, 21–26 Sept 1964: 820–2. (in Italian).
[13] L. Monolo, M. Pontiggia, A. Tarfani, A. Doriggi, Su di un caso di sparganosi cerebrale, Riv. Neurobiol. 22 (1976) 69–75 (in Italian).
[14] S. Pampiglione, M.I. Fioravanti, F. Rivasi, Human sparganosis in Italy. Case report and review of the European cases, APMIS 111 (2003) 349–354.
[15] Y.J.F. Garin, J. Frottier, A. Lavergne- Slove, R. Houdart, J.L. Poirot, Cutaneous sparganosis in France: the second case described from Europe. Case report, APMIS 105 (1) (1997) 14–16.
[16] G. Mougeot, M. Cambon, J.M. Menerath, V. DiMeglio, R. Houin, Human eye anterior chamber sparganosis, Parasite 6 (1999) 365–366.
[17] M. Rehák, L. Kolárová, I. Kohnová, J. Rehák, S. Mohlerová, E. Fric, O. Chrapek, Ocular sparganosis in the Czech Republic–a case report, Klin Mikrobiol Infekc Lek 12 (4) (2006) 161–164.
[18] F. Schauer, S. Poppert, K. Technau-Hafsi, M. Mockenhaupt, B. Muntau, G. Haecker, et al., Travel-aquired subcutaneous Sparganum proliferum infection diagnosed by molecular methods, Br. J. Dermatol. 170 (2014) 741–743.
[19] G. Bracaglia, S. Ranno, L. Mancinelli, M. Santoro, L. Cerroni, C. Massone, O. Sangueza, et al., A waterborn zoonotic helminthiase in an Italian diver: a case report of a cutaneous Sparganum infection and a review of European cases, Pathog. Global Health 109 (8) (2015) 383–386.
[20] V. Vortel, J. Pur, P. Halberstadt, J. Valkounová, Human sparganosis, Ceskoslovenska patologie 31 (1) (1995) 3–8.
[21] F. Botterel, P. Bouŕee, Ocular Sparganosis; a case report, J. Travel. Med. 10 (2003) 245–246.
[22] P. Walker, N.K. Cooper, A. Brandis, Cerebral sparganosis presenting as grand mal epilepsy, J. R. Army Med. Corps 153 (3) (2007) 189–190.
[23] D. Tappe, L. Berger, A. Haeupler, B. Muntau, P. Racz, Y. Harder, K. Specgt, et al., Case report: molecular diagnosis of subcutaneous *Spirometra erinaceieuropaei* sparganosis in Japanese immigrant, Am. J. Trop. Med. Hyg. 88 (1) (2013) 198–202.
[24] R.R. Gonzenbach, Y. Kong, B. Beck, A. Buck, M. Weller, A. Semmler, High-dose praziquantel therapy for cerebral sparganosis, J. Neurol. 260 (5) (2013) 1423–1425.
[25] H.M. Bennett, H.P. Mok, E. Grania- Klatosa, J.I. Tsai, E.J. Stanlej, N.M. Antum, et al., The genome of the sparganosis tapeworm *Spirometra erinaceieuropaei* isolated from the biopsy of a migrating brain lesion, Genome Biol. 15 (2014) 510–527.
[26] A. Lo Presti, D.T. Aguirre, P. De Andrés, L. Daoud, J. Fortes, J. Muñiz, Cerebral sparganosis: case report and review of the European cases, Acta Neurochir. 157 (8) (2015) 1339–1343.
[27] M. Kołodziej- Sobocińska, M. Tokarska, R. Kowalczyk, The first report of sparganosis (*Spirometra* sp.) in Euroasian badger (*Meles meles*), Parasitol. Int. 63 (2014) 397–399.
[28] M. Kołodziej- Sobocińska, M. Miniuk, I. Ruczyńska, M. Tokarska, Sparganosis in wild boar (*Sus scrofa*) - implicatons for veterinarians, hunters, and consumers, Vet. Parasitol. 227 (2016) 115–117.
[29] E. Kondzior, M. Tokarska, R. Kowalczyk, I. Ruczyńska, W. Sobociński, M. Kołodziej-Sobocińska, The first case of genetically confirmed sparganosis (*Spirometra erinaceieuropaei*) in European reptiles, Parasitol. Res. 117 (11) (2018) 3659–3662.
[30] S. Furmaga, *Spirometra janickii* sp. n. (Diphyllobothriidae), Acta Parasitol. Pol. 1 (1953) 1–59.
[31] A. Yoshida, K. Matsuo, J. Moribe, R. Tanaka, T. Kikuchi, E. Nagayasu, et al., Venison, another source of Paragonimus westermani infection, Parasitol. Int. 65 (6) (2016) 607–612.