A waterborn zoonotic helminthiasis in an Italian diver: a case report of a cutaneous *Sparganum* infection and a review of European cases

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Many waterborne helminthes are opportunistic parasites that can travel directly from animals to man and may contain forms capable of penetrating the skin. Among these, *Sparganum* is the pseudophyllidean tapeworm that belongs to the genus *Spirometra*, which is responsible for parasitic zoonosis; it is rarely detected in Europe and is caused by the plerocercoid infective larva. Thus far, only six cases of cutaneous and ocular sparganosis have been reported in Europe; two and four cases have occurred in France and Italy, respectively. Herein, we describe a new case of sparganosis in Italy that affected a male diver who presented to the Bambino Gesù Children's Hospital of Rome. The patient's skin biopsy was submitted to the Parasitology department who, in consultation with Pathology, concluded that the morphologic and microscopic findings were those of *Sparganum* spp. larvae. The patient recovered following a single dose of 600 mg praziquantel.

Keywords: Waterborne zoonotic helminthes *Sparganum*, *Spirometra*, Europe, Diver, Cutaneous infection

Introduction

Waterborne zoonotic helminthiasis are opportunistic parasites that can be transmitted from animals to man through water. The most significant zoonotic waterborne diseases are caused by snail-mediated and copepod-mediated helminthes transmitted by contaminated water.¹ Some helminthes, such as nematodes and cestodes, use one or more copepods as the intermediate host. Ingestion of these copepods and contact with contaminated water are two major risk factors for human infection. Sparganosis belongs to this group of waterborne diseases caused by a plerocercoid

tapeworm or second-stage larva *Sparganum* of the genus *Spirometra* (Pseudophyllidea: Diphyllbothriidae); the life cycle requires two intermediate hosts, a copepod and any one of a variety of vertebrates including fish, amphibians, reptiles, etc. Humans are accidental hosts. The infection is usually asymptomatic, and it is discovered accidentally, frequently during an unrelated surgical intervention. The larva can be found in different organs and, depending on the location, might produce serious damage, especially if it is located in the eyes,² heart,³ or brain⁴; however, it is generally located in the subcutaneous tissue.

Herein, we present an unexpected case of cutaneous sparganosis in an Italian male diver and review cases of cutaneous and ocular sparganosis that occurred in Europe previously published in the literature.

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Figure 1 A Papulopustular lesions within an erythematous plaque of about 2 cm on the left elbow; B After 1 month, growth of the plaque and erosions due to evolution of the pustular lesions are reported; C Inflammatory granulomatous dermal/subdermal process, haematoxylin and eosin (EE) staining, 40x; D After 3 months, almost complete healing of the lesion with mild residual erythema. Redness disappeared several weeks later.

Figure 2 A Section of *Sparganum* spp. larva, EE staining, 10x; B Excretory pores in *Sparganum* spp. section, PAS staining, 63x; C *Sparganum* spp. cycle.

Case Report

An Italian 56-year-old male diver was referred to our hospital in October 2013 after returning from a vacation in North Sardinia (Alghero). He reported a skin lesion on his left elbow that consisted of a 2-cm erythematous-squamous plaque that had progressively infiltrated; it had regular

edges and was covered by tense pustules (Figure 1A and B). The lesion was asymptomatic. The initial diagnoses were infection by *Leishmania* spp. or *Mycobacterium marinum*. However, these possibilities were ruled out by negative laboratory exams. There was no indication of immunosuppression.

Table 1 Summary of cases of human Sparganosis (cutaneous and ocular) from 1953 to 2014 described in Europe

Year	Source	Age/Sex	Symptoms	Lesion	Treatment	Locality	Infection route
1953	Pujatti	40, Female	Subcutaneous lump	Cutaneous, in the forearm	–	Italy	–
1953	Rossi and Genesi	28, Male	Subcutaneous lump	Cutaneous, in the inguinal region	–	Italy	Work in a rice field
1964	Bianchi	30, Male	Subcutaneous lump	Cutaneous, in the subclavian region	–	Italy	–
1997	Garin <i>et al.</i>	21, Male	Subcutaneous lump	Cutaneous, thoracic wall	None	France	Eating large amounts of smoked salmon on the Sabbath
1999	Mougeot <i>et al.</i>	14, Male	Eye pain	Conjunctival blood suffusion	–	France	–
2002	Pampiglione <i>et al.</i>	50, Male	Subcutaneous lump	Cutaneous lesion on thigh	–	Italy	–
2006	Rehák <i>et al.</i>	14, Male	Acute anterior uveitis	Ocular	Surgery	Czech Republic	Swallowed water
2014	Schauer <i>et al.</i>	61, Male	Persistent growth of lesion	Itchy reddish patch on right chest	Surgery	Germany	Fishing in white water

A punch skin biopsy was obtained and demonstrated an intense lympho-granulomatous dermal and hypodermic inflammatory process containing numerous neutrophils and eosinophils forming multifocal necrotic abscesses (Figure 1C). Periodic acid-Schiff (PAS) diastase, Giemsa and Ziehl–Neelsen stains were negative, excluding fungal, bacterial and protozoal infections. However, a foreign ovoid body of 180 μ with a cellulose-like coating and internal peripheral oval corpuscles (8–10 μ) without evidence of a reproductive or digestive tract was observed.

Histopathologically, the features of these materials were suggestive of a microcellular algae, and the initial diagnosis was that of a granulomatous dermatitis due to a foreign body (i.e. vegetal material). The specimen was submitted for review by a multidisciplinary team, which analyzed additional transverse sections and concluded that the organism corresponded to the cestode *Sparganum* larva. In addition, a PAS stain allowed identification of larva excretory pores of about 10 μ (Figure 2A and B). Three months after treatment, the lesion had almost healed completely with only residual mild erythema. The redness disappeared several weeks later (Figure 1D). The patient was treated with a single dose of 600 mg praziquantel with complete resolution of the lesion. The patient gave consent for publication of this case report and any associated images.

Discussion

Sparganosis is a rare parasitic infestation. Adult larva parasitizes Canidae and Felidae species.¹ However, the cycle includes passage through two intermediate hosts: (i) the first hosts are freshwater crustaceans; (ii) the second

intermediate hosts belong to different species of vertebrates (snakes, frogs and rodents)⁵ (Figure 2C). *Spargana* can live for about 30 years in the host. The disease is acquired by ingestion or contact with fresh infested water or direct contact with intermediate hosts. Humans are accidental and aberrant hosts. In fact, human sparganosis is often an overlooked and underreported disease.

Most of the approximately 450 documented human cases come from Asia^{6,7} and North America.⁸ A few cases have been reported in South America⁹ and in Africa.¹⁰ Clinically, sparganosis has a predilection for cutaneous involvement. In Europe, and especially in Italy, it is a very rare infection, and only 10 cases have been described in Europe, 5 of which were in Italy.^{11,12}

The first human case reported in Italy in 1953¹¹ involved a 40-year-old woman from Genoa, in whom the larva was located in the subcutaneous tissue of the right forearm. The second Italian case was a 28-year-old man from Vercelli who worked in a rice field and in whom the larva was detected subcutaneously in the right inguinal region. Other cases were reported in Northern Italy.¹¹ Pampiglione *et al.* described cases of sparganosis in Europe until 2003, and Lo Presti *et al.*¹² reported cerebral cases of sparganosis until 2015. An overview of all cases of human cutaneous and ocular sparganosis reported in Europe is listed in Table 1; however, we reviewed in detail only the following two cases of cutaneous and ocular sparganosis not previously described in a review format and reported in Europe up to 2015.

In 2006, a 14-year-old male was admitted to the Ophthalmologic Hospital in Olomouc with the diagnosis of acute ocular uveitis.¹³ A living parasite was detected and

surgically removed via paracentesis. Histopathological examination revealed the parasite *Sparganum* in the young larval stage. The authors suspect that the most probable source of viable parasites came from swallowed water containing infected crustaceans from a dam near the town of Olomouc in Moravia. A second case reported in Europe was in 2014 in a 61-year-old male who presented to the hospital after travelling in Bolivia, Brazil and Paraguay. Upon his return, he noted an itchy reddish patch on his right chest. During the following weeks, the patch grew in size, and the central part became dark and tumescent.¹⁴ A surgical excision was performed, and the helminth was extracted from the surrounding tissue. He had no impairment in his general condition and showed no sequelae or signs of infection. The source of infection remained unclear. In Italy, sparganosis is common in animals, especially in the districts of Turin, Padua, Pavia, Bologna and Cagliari, but it is rarely described in humans. Our case report is the seventh cutaneous case in Europe and the fifth Italian case. Herein, we describe a man who practised diving and water sports. The patient denied eating raw meat and fish and had no contacts with pets or exotic animals. The infection manifested as a lesion on his left elbow in October 2013 upon his return from holidays in North Sardinia (Alghero). Sections of *Sparganum* were carefully evaluated through analysis of both structural aspects and measurements, which led to a morphology-based diagnosis. The lack of sufficient DNA material hampered molecular diagnosis. It was inferred that the patient had been infected by swallowing contaminated water. Indeed, *Sparganum*, similar to *Gnathostoma*,¹⁵ can be acquired by eating raw fish or frogs or by water ingestion; considering that the patient did not have a history of eating such food, the second transmission hypothesis was assumed.

Many diseases can be caught while diving and swimming; those caused by organisms especially adapted to water environments can be both generalized and serious in nature. Although humans are accidental hosts, sparganosis can cause severe tissue or life-threatening damage. This case report is a warning for clinicians to keep parasitosis on the differential when examining a patient with a persistent skin lesion and suggests that divers in contaminated water need special clothing and protection as well as post-dive cleansing techniques. Further studies on endemic infected areas may be necessary to prevent further spread of these diseases.¹⁶ Screening exams to identify parasites such as sparganosis may be necessary.

Conflict of Interests

Authors have no conflict of interests.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the editor of this journal.

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