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Case report

Disseminated cysticercosis in a non-endemic area of Europe: diagnostic challenges and successful management of an unusual parasitic infection

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ABSTRACT. Cysticercosis caused by the larval forms of *Taenia solium* is observed sporadically in humans from non-endemic areas and is challenging to diagnose. We report an extraordinary case of 81-year-old woman from a rural region of Poland with worsening neurological symptoms suspected of brain malignancy. Imaging and serological tests confirmed disseminated cysticercosis with multiple space-occupying lesions located in the brain, soft tissues, and liver. Systemic cysticercosis in a white woman without a history of international travel is an exceptional clinical finding. Treatment with albendazole and corticosteroids led to significant reduction of pathological masses and clinical improvement. This case highlights the importance of considering unusual parasitic infections in neurological evaluations, even in non-endemic areas.

Keywords: disseminated cysticercosis, neurocysticercosis, *Taenia solium*, pork tapeworm, brain lesions, magnetic resonance, albendazole therapy

Introduction

Cysticercosis is a parasitic disease caused by the larvae of the tapeworm *Taenia solium* widely observed in developing countries in warm climate areas where pigs are raised. Infection most commonly occurs through the consumption of tapeworm eggs via the faecal-oral route. Humans become accidental intermediate hosts, and the tapeworm larvae can localize in almost all tissues and internal organs. In clinical practice, they are most commonly detected in the central nervous system because they then cause alarming and life-threatening symptoms and prompt further diagnostic workup [1].

The most common clinical manifestations of central nervous system involvement are seizures and in endemic regions, neurocysticercosis is responsible for up to 30% of all epilepsy cases [2]. The infection is considered to be the main cause of seizures in tropical areas, and recurrent seizures occur in about 80% of symptomatic cases of neurocysticercosis [2]. Depending on the location (intraparenchymal or extraparenchymal) and the number of cysts in the brain, various disease manifestations are possible – focal symptoms, signs of increased intracranial pressure, cerebrovascular complications (bleeding, strokes) and hydrocephalus including herniation [3].

Extraparenchymal localization – in the brain ventricles and subarachnoid space, is associated with a worse clinical prognosis [4]. Other clinical manifestations include cognitive decline, psychotic episodes, isolated headaches, involuntary movements, or eye damage. An important risk factor for the occurrence of cysticercosis in developed countries is travel to hyperendemic regions of the world, especially to Latin America, sub-Saharan Africa, Southeast Asia, as well as population migrations [5]. An increasing number of

immigrants from endemic regions constitutes an underestimated potential risk of infection in countries where local transmission is usually low.

Case report

We present a case of an 81-year-old woman who was referred to the clinical hospital for further investigation and management of lesions detected in brain following magnetic resonance imaging (MRI) and computed tomography (CT) scan. Imaging tests were ordered on the visit to the neurologist, during which she complained of worsening symptoms of Parkinson's disease. Additionally, new concerning symptoms have appeared, such as tremors, dizziness, balance disturbances, speech disorders, tingling, and weakness of the muscles in the left lower limb. The primary suspicion was of a malignant nature of the changes; therefore the patient was urgently referred to the neurological department of the Poznań University of Medical Sciences, Poland for a medical consultation.

The patient, living in a rural area with her family, is exposed to a long-term contact with pigs. She denies any foreign travel outside Poland, and does not recall any cases of tapeworm infection in her immediate surroundings. Her village had been known for years for breeding pigs and many generations of the family have always worked on the farm. She had never been diagnosed with intestinal taeniosis, and she never knew any other persons around her who had tapeworms or any significant abdominal problems. In the medical history, secondary parkinsonism, hypothyroidism, type II diabetes, and hypertension are documented. She underwent cataract surgery in 2022. Currently, she is taking acetylsalicylic acid, amlodipine with ramipril, atorvastatin, metformin, beta-histine, and levodopa with benserazide.

Upon admission to the Department of Neurology, the patient was in good general condition, with adequate cardiovascular and respiratory function. In the general physical examination, there were no significant deviations noted. Neurological examination revealed weakness in the muscles of the left lower limb (4.5 on the Lovett scale). During hospitalization, a repeat MRI was ordered. Following the radiologist's suggestion, a consultation with a specialist in clinical parasitology and tropical medicine was requested. The radiologist identified characteristic changes indicative of an infection with the larval form of *T. solium* – active hypodense

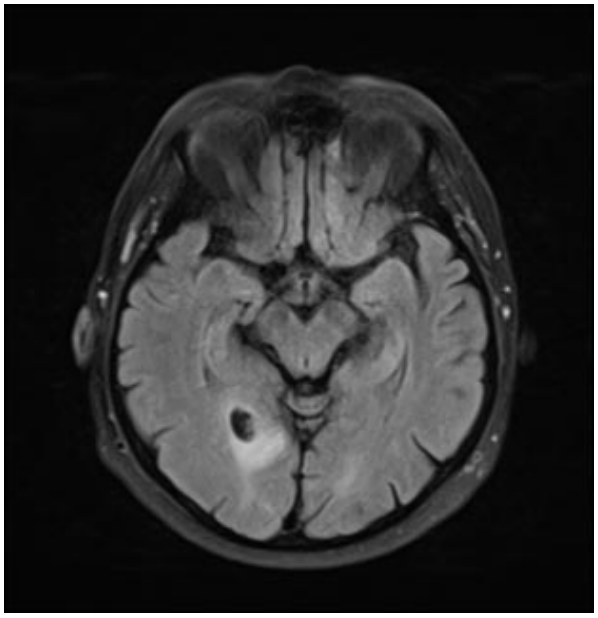

Fig. 1. Typical cysticercus of *T. solium* in the right occipital lobe of the brain with visible scolex as an internal nodule in a cyst (hole-with-dot sign) surrounded by oedema. Magnetic resonance imaging (MRI). The authors' collection.

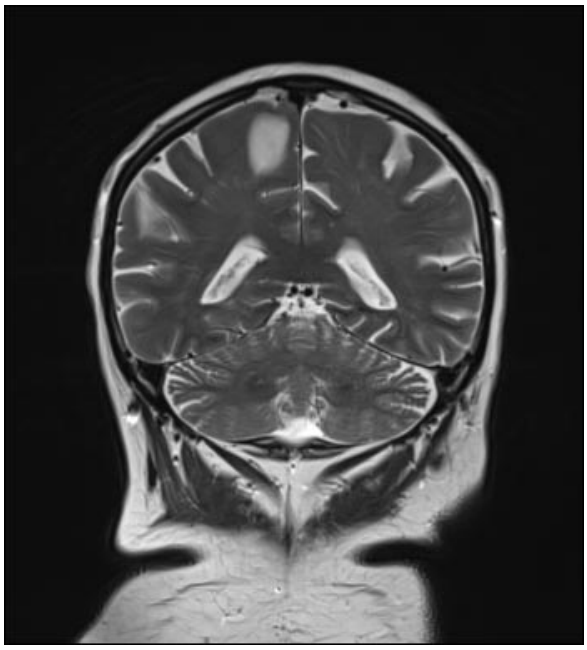

Fig. 2. Hyperdense cysticercus of *T. solium* in the brain's right parietal lobe. Magnetic resonance imaging (MRI). The authors' collection.

parenchymal cysts with internal echoes suggesting the presence of a tapeworm scolex as an internal asymmetric nodule inside (a picture of a larger black ball with a white tennis ball inside) through hyperdense or mixed lesions, and completely calcified forms. The two largest parenchymal lesions,

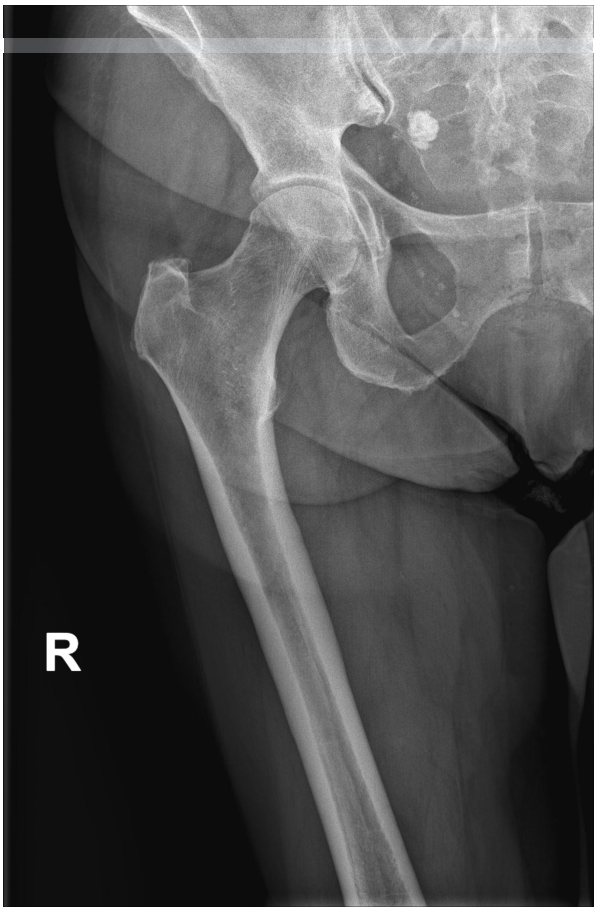

Fig. 3. Irregular calcified lesions located in the soft tissue of pelvis minor and right thigh corresponded to *T. solium* larvae. X-ray examination. The authors' collection.

measuring 22 × 19 mm and 28 × 15 mm, respectively, involved the right occipital lobe and the right parietal lobe (Fig. 1, Fig. 2).

The patient was urgently transferred to the Department of Tropical and Parasitic Diseases for a further clinical evaluation. Serological tests as ELISA (Bordier Affinity, Crisier, Switzerland) and Western-blot (LDBIO Diagnostics, Lyon, France) confirmed the presence of a specific IgG antibody against *T. solium* in serum samples. Due to the location of focal lesions in the brain, there were direct contraindications to lumbar puncture with examination of cerebrospinal fluid. Peripheral blood morphology with leucocyte smear show significant leukocytosis (17.08×10^9/L) with neutrophilia (10.98×10^9/L) with no changes in inflammatory markers. Biochemical tests revealed a slight elevation in the concentration of alanine transaminase (ALT) with a result of 54 IU/L (with a normal range of 10–31 U/L), aspartate transferase (AST) at a level of 38 IU/L (with a normal range of 10–31 U/L), and gamma-glutamyl transferase (GGT) at a level 37 U/L (with a normal range of 9–36 U/L). There was no hypereosinophilia or hypergammaglobulinemia in the peripheral blood. A concentration of phosphocreatine kinase (CPK) has been slightly elevated (215 U/L, with a normal range of 29–168 U/L) indicating a location of the parasitic infection also in peripheral muscles.

Suspecting a disseminated form of the disease, an X-ray examination of the femoral bones was ordered. In the soft tissue projection of the left thigh, three small opacities up to 3 mm were visualized, and in the projection of the right iliac bone, several circular opacities with a diameter of 3–4 mm were observed – most likely cysticerci (Fig. 3). In the abdominal ultrasound and computed tomography in segment V and VIII of the liver, longitudinal calcifications measuring 19 and 10 mm in length were observed, as well as 2 calcifications with a diameter of up to 5 mm which corresponded to inactive larval forms of *T. solium*. They presented classic "cigarette" forms of the larvae (Fig. 4). There were no pathological changes on fundoscopic examination, optical coherence tomography (OCT) of the eyes and echocardiography. The stool examination did not reveal any eggs of *Taenia* spp. or other intestinal parasites.

On a basis of typical risk factors from epidemiological interview, as well as imaging and serological data, the final diagnosis of systemic cysticercosis has been established. A 14-day course of albendazole therapy (400 mg per os twice a day) was initiated, supplemented with methyl-prednisolone (24 mg in a single dose), aiming to

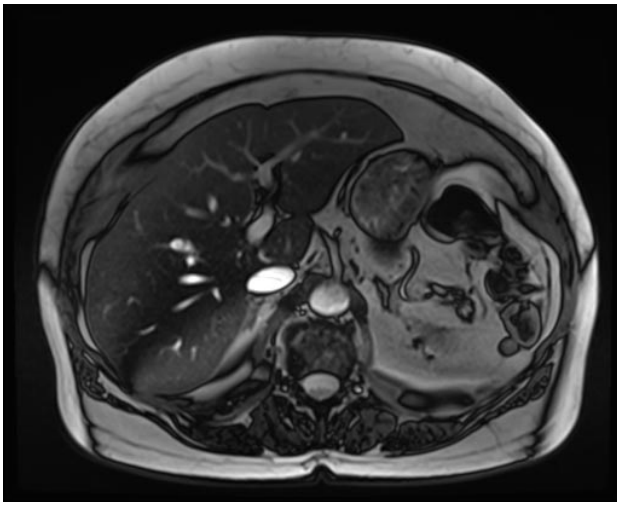

Fig. 4. Multiple longitudinal cigarette-like lesions discovered in the liver. Magnetic resonance imaging (MRI). The authors' collection.

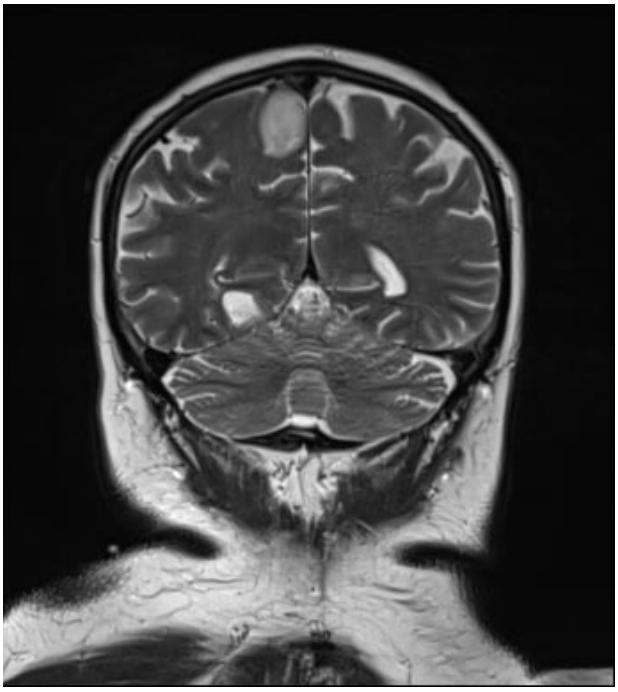

Fig. 5. Degenerative hyperdense larval forms of *T. solium* surrounded by gliosis in the brain 6 months after treatment. Magnetic resonance imaging (MRI). The authors' collection.

prevent brain edema and reduce perilesional inflammation, which can be caused by rapid antigen release during the degradation of tapeworm larvae.

After treatment, the patient reported significant improvement in neurological symptoms, muscle strength, and motor function, and was discharged home in good general condition. A 6-month follow-up documented successful clinical outcome without persistence of focal neurological deficits.

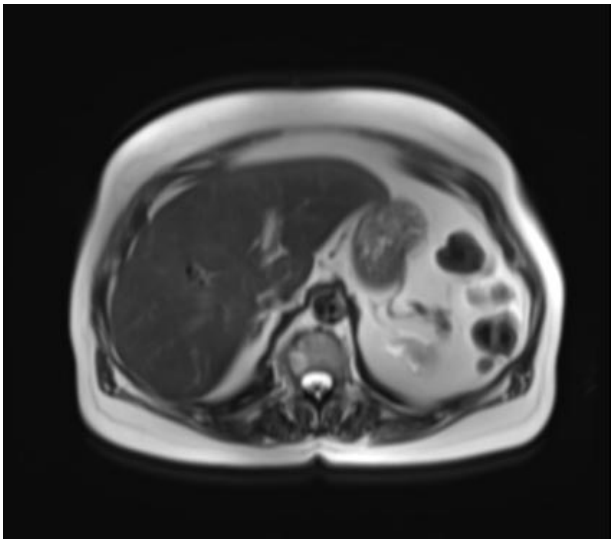

Fig. 6. Two irregular hyperdense lesions observed in the liver. Magnetic resonance imaging (MRI). The authors' collection.

In the follow-up laboratory test results, a decrease in the level of ALT (36 U/L, with a normal range of 10–31 U/L) AST (29 U/L, with a normal range of 10–31 U/L), GGTP (25 U/L, with a normal range of 9–36 U/L) and CPK (73 U/L, with a normal range of 29–168 U/L) was noted. Apart from that, no clinically significant deviations were observed.

In the follow-up MRI of the head performed 6 months later on, the space-occupying lesions were found to be more consolidated and inactive with a size of 28 × 15 mm in the parietal lobe and 20 × 18 mm in the occipital lobe of the brain (Fig. 5). Based on this, it is concluded that these changes are not of neoplastic origin, which confirms the accuracy of the diagnosis.

In the abdominal MRI examination, two hyperdense lesions measuring 11 mm and 6 mm were detected in segments VII and VI of the liver. This image indicates amorphous calcifications, which may occur in the course of cysticercosis (Fig. 6). A repeat abdominal ultrasound confirmed the findings obtained in the MRI and showed stability of the lesions compared to those visualized in the previous examination.

Discussion

We are describing a very rare and unusual indigenous case of a Polish patient with generalized cysticercosis. We revealed tapeworm larvae in the central nervous system, soft tissues of the abdomen and thigh, and additionally in the liver – an amazing location not previously visualized in the imaging studies according to the actual database. In hyperendemic areas of Central and South America, sub-Saharan Africa or Asia, systemic or neurological infection due to *T. solium* is quite frequently diagnosed but autochthonous disseminated case from Poland is a spectacular observation.

Systemic cysticercosis with innumerable *T. solium* larvae observed in skeletal muscles, subcutaneous tissue, tongue, skin, heart, central nervous system, retina, parotid gland has been already documented in non-vegetarian native people, including children, from India, Nepal, Vietnam, Mexico, Peru, and in immigrants from India, Ecuador, Bolivia, Guatemala, Nicaragua, Cape Verde, Angola, Mozambique, Guinea-Bissau, Sao Tome and Principe Islands or travellers to Africa, South Korea or China [6–12].

The clinical and imaging diagnosis is more suggestive and clearer when multiple firm, well-circumscribed, non-tender subcutaneous nodules are present throughout the body during clinical examination, along with innumerable tiny, ring-shaped, nodular enhancing lesions in the brain, displaying a highly pathognomonic „starry sky" appearance on magnetic resonance imaging [5,13–16]. The classic spreading of cysticerci to muscular or subcutaneous locations may remain completely asymptomatic or may imitate benign tumors such as lipomas or fibromas. In suspected cases, fine-needle aspiration biopsy or histopathological section is essential in establishing the final identification of the parasite [17]. Recently, whole body – magnetic resonance imaging (WB-MRI) using the total imaging matrix coil has been shown as a valuable stand-alone modality in the diagnosis and management of disseminate cysticercosis [18].

A location of *T. solium* larvae in the central nervous system is a diagnostic feature but may suggest any neurological disease. Cysticercosis can remain undetected for years until the first neurological symptoms appear. In described case, the evidence of characteristic cystic lesions showing the scolex on neuroimaging examinations has been an absolute diagnostic criterion of the disease. Resolution of intracranial cystic lesions after therapy with albendazole is considered to be a major clinical sign. The detection of cysticerci outside the central nervous system together with epidemiological factors complete a list of definitive markers which are helpful in a diagnostic certainty [19].

In tropical areas of equatorial Africa, the differential diagnosis with *Onchocerca volvulus* nodules (onchocercomata) containing adult filariae is highly recommended. On the other hand, post kala-azar dermal leishmaniosis which is endemic in many parts of India, Nepal, Bangladesh, Sudan, Ethiopia and Kenia that is seen in untreated or inadequately treated cases of visceral leishmaniosis should be included into the differential diagnosis. *Mycobacterium leprae* infection, including the most severe lepromatous form or atypical Mycobacteria can also imitate a similar clinical picture and give some significant diagnostic difficulties or delay in the identification and treatment.

In accordance with the consensus in the current literature, we administered albendazole in two divided doses of 400 mg for 14 days. Albendazole monotherapy has been shown to be more effective compared to praziquantel, with lower organ toxicity [5]. In some cases, a combination therapy of albendazole and praziquantel might be used. Glucocorticosteroid coverage was used during antiparasitic treatment [5]. Due to the absence of epileptic symptoms, we decided not to administer antiepileptic medications [3,21].

In the described case, we encountered several diagnostic challenges. One of them was the coexistence of neurological conditions, through which the patient incorrectly interpreted the initial symptoms as an exacerbation of Parkinson's disease. Additionally, no epileptic seizures, which are the most common and characteristic symptoms of neurocysticercosis, were observed in her case [5]. Furthermore, patient's absence of a foreign travel history was noted, which often excludes cysticercosis from the differential diagnosis [18,22].

Breeding domestic pigs is a substantial risk factor for the transmission of *T. solium* and the contracting of the intestinal infection by humans. This is subsequently indicative of the potential presence of the parasite eggs in the rural environment if local populations are infected [5,22]. In our case, we can conclude that it was one of the key causes of the infection. The patient independently ran a pig farm, and in the neighborhood, there is a large industrial pig farm. The route of infection remains an open question. Considerations include: autoinoculation, transmission of eggs from individuals in close proximity with undiagnosed tapeworm infection, and the faecal-oral route due to the lack of proper sanitary conditions in the farm environment. On the other hand, long life expectancy favored high and repeated exposure to the infection in a potentially permanently contaminated environment by a direct contact with a high inoculum of *T. solium* eggs. It remains a mystery why no other cases of the disease have been observed in the same geographical area. The disease should show a family occurrence or cause occupational outbreaks. There is also a possibility that a senior patient will be less likely to follow hygiene rules on the farm. Generalized cysticercosis with thousands of calcifications due to haematogenous spreading of cysticerci with the characteristic "rice grain" pattern has been documented in a similar in age 98-year-old woman from a nursing facility in Portugal [20]. Moreover, intestinal taeniosis preceding cysticercosis with a passive excretion of short tapeworm segments in the feces may have gone unnoticed by the elderly

patient. For these reasons, it seems to be highly advisable to perform screening tests for a local population, for example, determination of *T. solium* – specific IgG antibody levels in peripheral blood or a non-invasive soft tissue X-ray for a presence of degenerating larval forms of the parasite, as well as a direct microscopic examination of stool samples for a presence of mature proglottids of the adult tapeworm or *Taenia* spp. eggs. A detection of specific IgG antibodies against glycoproteins of 50, 39–42, 24, 21, 18, 14 or 13 kDa in the patient's blood or cerebrospinal fluid by using immunoblotting (Western-blot) is characterized by a high sensitivity of 98% and specificity of 100%, and is recommended to be used for a final confirmation of cysticercosis in non-endemic areas [23].

Neurocysticercosis, rare in our geographic region, poses a diagnostic challenge due to its nonspecific symptoms or signs. Larval forms of the parasite located in subcutaneous tissue or muscles usually do not cause clinical manifestations and remain unrecognized. A lack of widespread awareness of this disease among clinicians and radiologists can lead to delays in its diagnosis and treatment, therefore, it is crucial to consider this as a potential cause of focal neurological symptoms.

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