

## Case Report

## Cerebral mass in a 13-year-old girl following long-term sojourn in the Tropics

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Cysticercosis of the central nervous system is the main cause of late-onset epilepsy in tropical countries. The case of a 13-year-old German girl with a generalized seizure following long-term sojourns in the Tropics is reported. Cranial imaging showed two cerebral lesions with central calcifications. Serological, molecular and cultural examination of cerebrospinal fluid and blood was negative for various parasites, fungi and bacteria including mycobacteria. Histopathological examination after neurosurgical resection revealed calcareous bodies pathognomonic for platyhelminths, in particular tapeworms. Taken together, the radiological and histopathological findings indicate infection with cysticerci, the larvae of *Taenia solium*.

## Introduction

Neurocysticercosis is the most common parasitic infection of the central nervous system (Carpio, 2002). It is caused by the larval stage of the tapeworm *Taenia solium*. Human cases in Europe including Germany are particularly rare (Wiegand *et al.*, 1999). However, in tropical countries, especially in South America, it has been shown to be the main cause of late-onset epilepsy (Rosenfeld, 2003).

Pigs are intermediate hosts of *Taenia solium* larvae (metacystode, *Cysticercus cellulosae*). Humans are definitive hosts and may develop enteral taeniosis after consumption of undercooked pork. In the human intestine cysticerci mature to the adult tapeworm. In contrast, cysticercosis is acquired by ingestion of *Taenia solium* eggs present in faeces-contaminated food. In the intestine, the larvae (oncosphere) hatch, penetrate the gut wall and are transported via the bloodstream to neural, dermal and muscular tissues. Here, the cysticercus develops. In this case, humans serve as intermediate but dead-end hosts.

This tropism accounts for the clinical presentation. In the central nervous system, focal epilepsy is most common, in addition to personality changes and cognitive deficits. If cerebrospinal fluid (CSF) circulation is impaired due to multiple, space-occupying lesions or cysticerci in the subarachnoid space, hydrocephalus may result. In children, most commonly a solitary parenchymal lesion can be found; however, encephalitis may be present with large numbers of cysticerci (Rosenfeld, 2003). Autoinfections, where humans

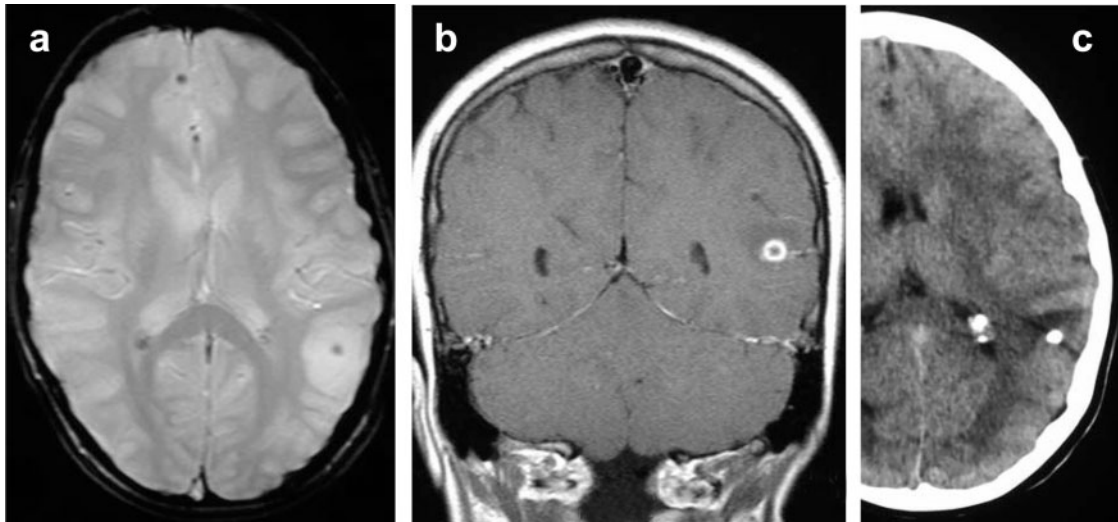
carry both the adult tapeworm in their intestine and cysticerci, occur in 5–40% of carriers. The tapeworm can shed as many as 300 000 eggs daily (Carpio, 2002).

Diagnosis of cysticercosis is based on serology and radiological imaging. A carrier state of adult *Taenia solium* can easily be detected by stool examination for ova and proglottids. Anthelmintic treatment of cerebral cysticercosis consists of albendazole, praziquantel, corticosteroids and anti-epileptic drugs (Carpio, 2002; Hawk *et al.*, 2005; Rosenfeld, 2003). In severe cases, surgery and CSF shunting may be required.

## Case report

A 13-year-old German girl who grew up in different countries of South America (among others, Uruguay, 5 years) and South East Asia (among others, Thailand, 2 years) presented with a generalized seizure and a history of aphasia. The neurological examination was otherwise normal. There was no fever. Total blood count, white blood count and serum electrophoresis were unremarkable. CRP and IgE levels were within normal limits. The electroencephalography showed a slow wave focus of the left temporoparietal cortex. A corresponding lesion could be seen in magnetic resonance imaging (MRI) (Fig. 1a, b) and cranial computed tomography (CT). CT showed a calcified lesion (Fig. 1c). On the MRI, the lesion was surrounded by a ring of gadolinium enhancement and perifocal oedema (Fig. 1a, b). Another plain calcification was noted in the right frontal lobe (not shown). Because of a positive Mendel–Mantoux test, cerebral tuberculoma had to be considered as differential diagnosis to parasitosis. The chest X-ray showed no abnormalities. Furthermore, in gastric

Abbreviations: CSF, cerebrospinal fluid; CT, computed tomography; MRI, magnetic resonance imaging.



**Fig. 1.** Cranial imaging. (a) Proton-weighted and (b) T1-weighted (+ i.v. gadolinium contrast dye) cranial MRI; (c) cranial CT scan of the brain showing a calcified mass in the left temporoparietal lobe.

fluid, sputum, and material obtained from the later performed intraoperative bronchoalveolar lavage, no mycobacteria could be detected. In CSF, cell count and protein and glucose concentrations were normal. In addition, Gram and Ziehl–Neelsen stains were negative. CSF culture yielded no growth of eubacteria, mycobacteria or fungi. A universal PCR detecting eubacteria and the *Mycobacterium tuberculosis* complex was negative.

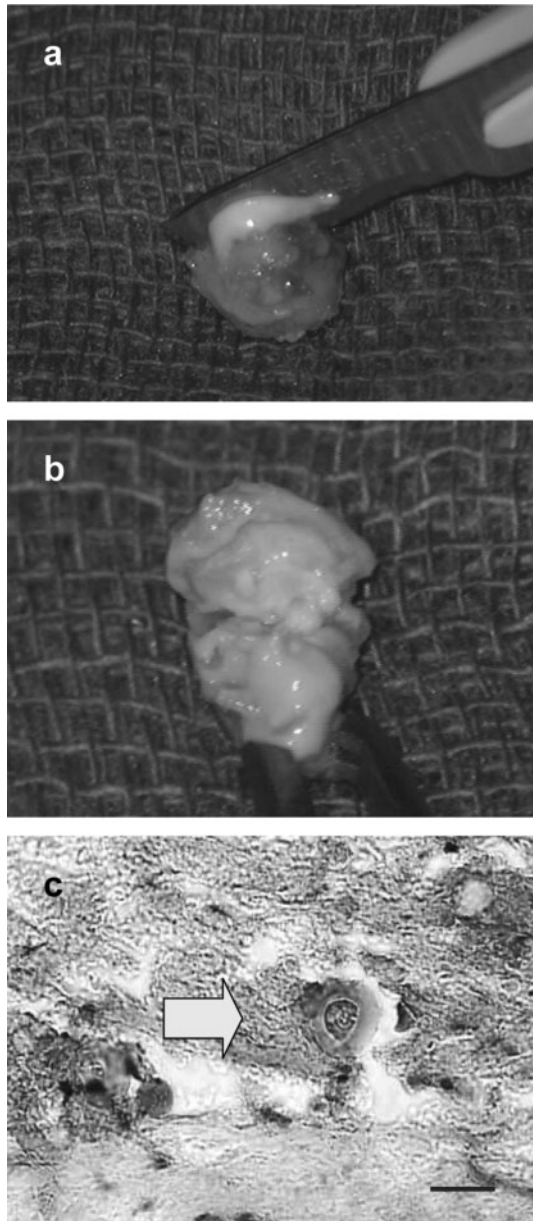
Serology of CSF and blood for various fungi (*Candida*, *Aspergillus*, *Cryptococcus*, *Histoplasma* and *Coccidioides*) and parasites (*Toxoplasma*, *Echinococcus*, *Taenia solium* cysticerci, *Toxocara*, *Trichinella*, *Dirofilaria*, *Paragonimus*, *Fasciola* and *Schistosoma*) was unremarkable. Because of the rather superficial localization and the unknown aetiology, complete neurosurgical resection of an encapsulated structure of 6 mm diameter (Fig. 2a, b) was performed. Perioperatively, an antimycobacterial and antihelminthic treatment using streptomycin, rifampicin, isoniazid, albendazole (400 mg b.i.d.) and dexamethasone (0.45 mg kg<sup>-1</sup> per day) was started. Both treatment strategies were chosen, because at that time none of the considered infections including tuberculosis could be excluded completely. Finally, histopathological examination revealed extensive tissue necrosis, calcification and sparsely calcareous bodies pathognomonic for platyhelminths, in particular tapeworms (Fig. 2c). Stool examination for ova and proglottids and helminth larvae was negative. Because mycobacterial PCR and microscopy of the excised tissue and a bronchoalveolar lavage were negative, antimycobacterial therapy was reduced to isoniazid treatment only, scheduled to last 9 months. Six weeks later, mycobacterial cultures were still negative. Albendazole treatment together with dexamethasone was continued for 1 week after surgery and was well tolerated. No further neurological events were reported during 1 year of follow-up.

## Discussion

Cysticercosis due to *Taenia solium* larvae is the most commonly encountered parasitic infection of the brain. Viable cysticerci show only little surrounding inflammation and are often asymptomatic. When the parasite degenerates, inflammation mediated by immunological effector cells leads to development of clinical symptoms. Even a long time after calcification, a renewed inflammatory response may be evoked causing surrounding oedema and seizures (Carpio, 2002), as presented in our case. Serology is regarded to be highly sensitive and specific for the diagnosis of cysticercosis. However, sensitivity decreases from >90% to <25% when only one lesion is present and may become completely negative after the cysticercus dies and calcifies (Garcia *et al.*, 1997; Wilson *et al.*, 1991). Repeated immunoblots for cysticercosis were negative in the case depicted here. When only a single lesion is present serology is negative in 72% of cases. Thus, negative serology does not rule out cysticercosis (Wilson *et al.*, 1991).

No data on the seroprevalence in Uruguay exist. However, in Brazil seroepidemiological studies showed a prevalence of 0.68–5.2% (Agapejev, 1996). In Thailand, 20 of 972 patients presenting with epilepsy were diagnosed with neurocysticercosis (Yodnopaklow & Mahuntussanapong, 2000), but no data on the seroprevalence are available. The seroprevalence in Vietnam is 5–7% (Rajshkhar *et al.*, 2003), and similar data can be expected for Thailand.

Radiological imaging is the preferred method for diagnosing cerebral infestations of cysticercosis, but may be difficult to interpret, if very few and already calcified lesions are visible. Eventually, a neurosurgical approach can aid in clarifying the aetiology. In the case described, diagnosis was accomplished by the histopathological finding of calcareous bodies



**Fig. 2.** Anatomy and microscopy of the tissue lesion. (a, b) Excised tissue from the left temporoparietal lobe. The cut surface revealed a non-caseating, soft and gelatinous consistency. (c) Histology of the calcified brain lesion (haematoxylin and eosin staining; bar, 200  $\mu$ m).

in the necrotic cerebral lesion. However, calcareous bodies can be found in all platyhelminths (cestodes and trematodes). Their function is only poorly understood, but accumulation can be found in host tissues surrounding parasites (Etges & Marinakis, 1991). Other cerebral infections by metacestodes, such as various forms of echinococcosis (due to *Echinococcus granulosus*, *Echinococcus*

*multilocularis*, *Echinococcus vogeli* and *Echinococcus oligarthus*), coenurosis (caused by *Multiceps multiceps*) and, rarely, sparganosis (caused by *Spirometra mansoni*), must be considered, but appear distinctly on MRI and CT (Cummings *et al.*, 2000; Ing *et al.*, 1998; Tuzun *et al.*, 2004). With regard to trematodes, ectopic localizations of *Schistosoma* eggs and juveniles of *Fasciola* and *Paragonimus* should also be taken into account. Infection should be excluded by serology, stool, urine and sputum examination.

In conclusion, a parasitosis of the central nervous system, in particular neurocysticercosis, must be considered in patients with new-onset epilepsy after returning from a long-term stay in the Tropics, including immigrants.

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