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LETTER TO THE EDITORS

High-dose praziquantel therapy for cerebral sparganosis

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Dear Sirs,

A 39-year-old Bangladeshi man was admitted to our hospital with a first generalized epileptic seizure. Brain MRI showed multiple ring-enhancing lesions with perifocal edema (Fig. 1a and g, February 2008). CSF analysis showed 8 cells/µl and normal protein, glucose and lactate levels. Serum and CSF serological tests were negative for a broad range of parasites, fungi and bacteria.

The patient was suspected to have neurocysticercosis and was treated with albendazole (400 mg bi-daily for 30 days). However, follow up MRI showed new, adjacent lesions with ring- and tunnel-shaped enhancement; and punctate calcifications in the left parieto-occipital area in the CT scan (Fig. 1b, h and m, December 2009), suggesting that the treatment had been ineffective. We therefore performed a brain biopsy (with prior MRI for the localization of the sparganum, see Fig. 1c and i, February 2010), which revealed a degenerated cyst containing membrane-shaped structures with multiple foci of calcification. PCR was positive for *Spirometra erinaceieuropaei*, leading to the diagnosis of cerebral sparganosis.

Surgical removal of the parasite is the treatment of choice as standard anti-helmintic therapy is considered ineffective in sparganosis [1–3]. The repeated MRI indicated that the lesion had been wandering again (Fig. 1d and j). An ethylcholine PET showed a defined region with increased tracer uptake, most likely indicating the current location of the sparganum (Fig. 1n). However, the patient refused the recommended stereotactic surgery. We therefore treated the patient with a high-dose regimen of praziquantel (3 × 25 mg/kg body weight daily) for 7 days combined with cimetidine (3 × 400 mg daily) and a high carbohydrate diet to increase plasma levels of praziquantel [4]. The treatment was well-tolerated.

Follow-up MRIs 3 and 11 months later showed drastic improvement and the ethylcholine PET had normalized (Fig. 1e, f, k, l, o and p). Anti-sparganum antibody levels were determined from preserved CSF and serum samples by a specific ELISA [5]. Before therapy (Fig. 1, December 2009), anti-sparganum antibody levels were elevated in the serum (0.29) and in the CSF (0.55 normal value in serum and CSF < 0.22). After therapy (Fig. 1, June 2011), antibody levels had dropped to 0.09 in serum (a lumbar puncture was not repeated after therapy). Seizures discontinued and anticonvulsive treatment was slowly tapered.

Sparganosis is a rare parasitic disease caused by infestation with the larval cestode of *Spirometra* spp. Most cases have been reported in Southeast and Eastern Asia. Humans are infected by drinking unfiltered, contaminated water or through consumption of raw snakes and frogs. When the parasite invades the central nervous system, epileptic seizures and headache are common symptoms [2]. Characteristic MRI findings of cerebral sparganosis include a

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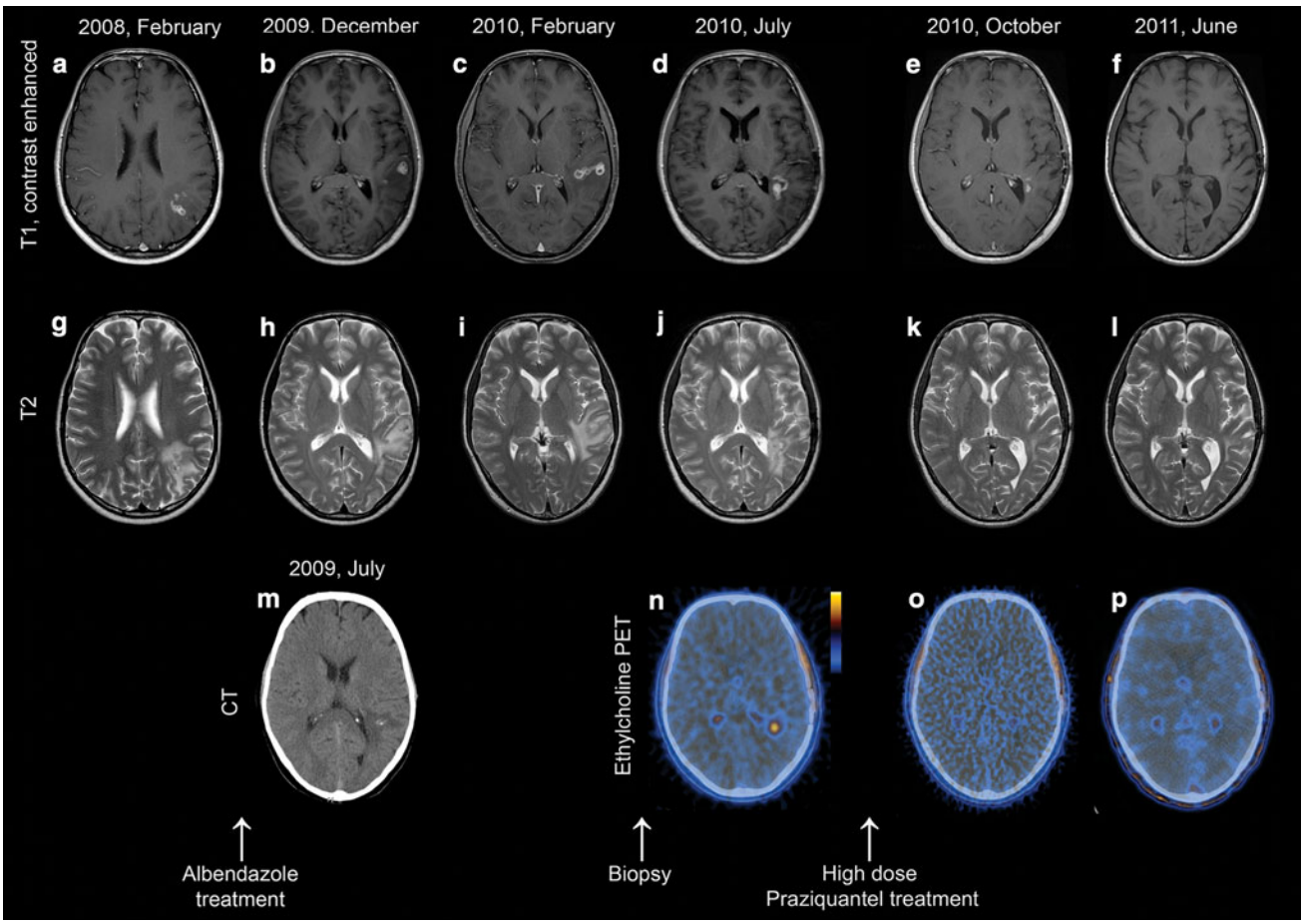

Fig. 1 Radiology findings. The *upper* and *middle row* show axial brain MRIs in chronological order. The *upper row* shows contrast-enhanced horizontal T1-weighted sections (**a–f**). The *middle row* shows the corresponding T2 sections (**g–l**). Characteristic punctate calcifications in the CT in the affected region are shown in (**m**). Repeated ethylcholine PET scans before and 3 and 11 months after praziquantel therapy are shown in (**n–p**). The relative amount of tracer uptake is *color-coded* with arbitrary units

'tunnel sign', and 'wandering lesions' on successive images representing the track of motion of the parasite [6, 7].

The treatment of choice for cerebral sparganosis is surgical removal because treatment with anti-helminthics, including praziquantel, has been described as not effective [1–3]. In our patient the clinical improvement, and the normalization of brain imaging and of anti-sparganum antibody levels suggest that the parasite was eradicated by high-dose praziquantel therapy. Although the close temporal correlation of therapy and presumed death of the parasite suggests a causal relationship we cannot rule out the possibility that this was, in fact, due to a coinciding, natural death of the parasite and not due to the chemotherapeutics. There is no information regarding the cestode's life expectancy in human tissue in the literature.

Cerebral sparganosis should be suspected in patients from endemic areas with indications of cerebral parasitic infection and "wandering lesions" in the MRI. High-dose praziquantel treatment may be considered in inoperable cases of cerebral sparganosis. Ethylcholine PET can be used preoperatively for the precise localization of the sparganum. This should be particularly valuable if the precise location is not known due to the often multifocal lesions in MRI.

Conflicts of interest On behalf of all authors, the corresponding author declares no conflict of interest.

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