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Case Report

Parenchymatous cerebral neurocysticercosis in a quadriplegic patient

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Objective: To present and discuss a case of cerebral neurocysticercosis in a quadriplegic patient.
Design: Case report of a case of neurocysticercosis in a high level spinal cord injury (SCI) patient who developed episodes of autonomic dysreflexia and orthostatic hypotension associated with transient neurologic deficits and seizures.
Setting: Spinal Cord Unit of the University Hospital of Geneva, Switzerland.
Subject: Single patient case report.
Main outcome measure: Clinical and radiological magnetic resonance imaging follow-up of the patient between July 1995 and October 1997.
Results: Treatment of cysticercosis with praziquantel relieved the patient from autonomic dysreflexia, symptomatic orthostatic hypotension, transitory neurological deficits and seizures.
Conclusion: Diagnosis of neurocysticercosis in a quadriplegic patient might be difficult because of frequent overlaps with some usual symptoms occurring in high level SCI, mostly autonomic dysreflexia and orthostatic hypotension. Neurocysticercosis should be kept in mind when a SCI patient living in, or coming from endemic zones presents with new neurological abnormalities and seizures. Magnetic resonance imaging appears to be more sensitive than computerised tomography to confirm the diagnosis of active cysticercosis. Treatment with praziquantel associated with cimetidine to increase the drug bioavailability and prednisone to reduce the inflammatory reaction gives good results.

Keywords: spinal cord injuries; cysticercosis; autonomic nervous system diseases

Introduction

In the United States, the national incidence of SCI varies between 7000 and 10 000 persons per year, with a prevalence of 150 000 to 200 000, quadriplegic accounting for approximately 55% of the cases.[1] Autonomic dysreflexia (AD) results from various noxious stimuli and occurs after the initial phase of spinal shock when the lesion is at or above the level of the sixth thoracic vertebra, that is above the major splanchnic outflow.[2,3] AD is often preventable and must be considered as an emergency as the sudden and severe rise in blood pressure may result in stroke, seizures and even death.[2,4] As sympathetic inhibitory impulses which originate above Th 6 are blocked as a consequence of injury, there is a relatively unrestricted sympathetic splanchnic outflow (Th 6–L2) below this level stimulated by intact sensory nerves with a release of norepinephrine, dopamine and dopamine-beta-hydroxylase. Release of these chemicals may cause pilo-erection, skin pallor and severe arterial vasoconstriction with sudden elevation of blood pressure frequently associated with headache. Episodes of AD are often associated with bradycardia, vasodilatation profuse sweating and skin flushing above the level of injury due to increased parasympathetic and sympathetic inhibitory compensatory mechanisms.

Another frequent problem encountered by patients with high level SCI is the reduced tolerance to the upright position. Orthostatic hypotension (OH) occurs from the deprivation of the sympathetic response that prevents pooling of blood in dependent parts and from the poor tissue turgor which allows extravasation of fluid. In most cases, this problem is of a temporary nature. For a small number of patients, OH persists and requires mechanical or pharmaceutical control.

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Taenia solium, the agent responsible for neurocysticercosis, is a very prevalent human parasite commonly found in the Slavic countries, Mexico and Latin America as well as in the Indian peninsula and China. In those regions, up to 33% of patients with seizures have antibodies to T. solium, compared with 2–11% prevalence in the general population, suggesting the likelihood that neurocysticercosis is an underlying cause of their disease.[5] Autopsy data from Mexico City show that the prevalence rate of neurocysticercosis ranges from 1.4 to 3.6% in the general population.[6] The relative frequency of high SCI and neurocysticercosis is at the best of our knowledge unknown so far.

Case report

A 52-year-old French white male patient presented with a complete C4 quadriplegia (Asia impairment scale = A) in July 1995 following a fracture with dislocation of C5 due to a motor car accident. Working for an international organisation, he had often been travelling in developing countries and had settled in Bolivia for the last ten years. The past medical history was irrelevant except for an episode of diarrhoea, more than ten years ago, related to a 'Taenia' worm while travelling in Africa. Urodynamics revealed an atonic bladder. Bladder drainage was by intermittent catheterisation. His blood pressure was 110/60 mmHg and pulse rate was 74/minute.

One month after onset of SCI, the patient presented with frequent episodes of OH responding moderately to etilefrine HCl and few episodes of AD which were attributed to bladder over-distension, a small sacral decubitus ulcer and a perianal abscess. Between October 5 and November 27, AD became more frequent, and the patient was often found confused. One episode was associated with transient aphasia, another with transient left facial palsy and the last with transient aphasia and right homonymous hemianopsia. Clinical examination was irrelevant apart for a mild splenomegaly and the transitory neurological deficits. Blood analysis was normal except for a mild normochromic normocytic anaemia. An electroencephalographic examination (EEG) done on November 20 showed abnormalities in the left temporal lobe. Three successive enhanced cerebral computerised tomography (CT) scans with 8 mm thickness contiguous sections were performed and described as normal showing no space occupying, haemorrhagic or ischaemic lesions and no signs of meningitis or encephalitis. A first 8 mm thick contiguous sections magnetic resonance imaging (MRI) examination was performed on December 8, 4 days after the last CT. This examination was able to show five lesions compatible with cysticerci and one granulomatous lesion in the left hemisphere (Figure 1). By the time the diagnosis was made, both enzyme-linked immunoelectrotransfer blot assay (EITB) and enzyme-linked immunosorbent assay (ELISA) showed positive immune reaction against cysticerci in blood serum but not in the cerebrospinal fluid which otherwise showed no protein or glucose alteration and no pleocytosis. Direct stool examinations showed no eggs or proglottids of the cestode. By February 1996, a second MRI showed a total of seven lesions at cystic or granulomatous stages and varying from 3–5 mm in diameter. The focal neurological manifestations correlated well with the location of these cysts.

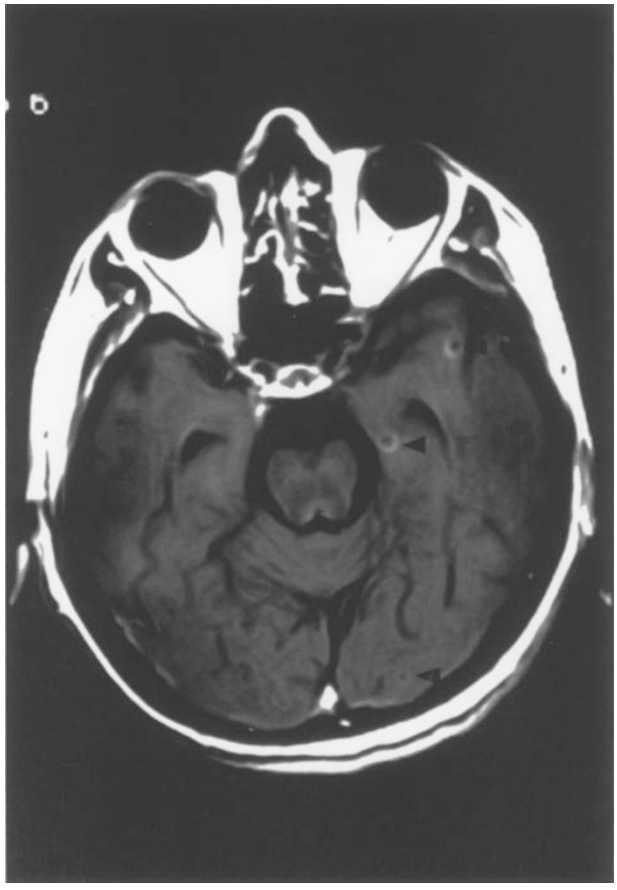

Figure 1 T1-weighted MRI images of the brain enhanced with gadolinum taken with TR 686 msec and TE 20 msec in the axial plane before therapy showing 3 ring-like enhancing live cystic lesions with scolex

Therapy consisted in a 3 weeks treatment with praziquantel at the dose of 25 mg/kg body-weight twice daily. Simultaneously, the patient received 1600 mg of cimetidine, 40 mg of prednisone and 2 mg of clonazepam daily. By May 1996, the patient was totally free of any symptoms and he could leave the hospital 5 months later without any anti-epileptic medication.

When the patient was contacted by telephone in October 1997, 1 year after discharge from hospital, we learned that he was doing well and that he has recovered partial sensation and movement in his thumbs. He has no more presented AD, epilepsy, confusional state or neurological deficits.

Spinal cysticercosis was considered but not investigated since the cause of SCI was well-defined.

Discussion

The diagnosis of neurocysticercosis in this quadriplegic patient was not easy as seizures, headaches, nausea, somnolence, speech disturbances and alterations in cognitive abilities can be encountered in various situations such as hypertensive encephalopathy, autonomic dysreflexia, orthostatic hypotension, ischaemic brain disease and space occupying lesions. Initially, blood pressure fluctuations and the transient neurologic deficits presented by the patient were attributed to AD which could have been stimulated by occasional bladder over-distension, the presence of a small decubitus ulcer and a perianal fistula. Dizziness and fainting were attributed to OH and the patient responded relatively well to specific measures such as compression stockings and etilefrine. Other possible aetiologies for his symptoms were not addressed before he presented with episodes of focal neurological deficits associated with confusion and disorientation. Potential embolic sources were ruled out by cardiac echography and a Doppler-echographic examination of the carotid trunks. By the end of November, non-convulsive epileptic seizures were suspected. EEG showed an irritative focus in the left temporal lobe and the positive diagnosis of neurocysticercosis was made on the basis of the MRI. Although the CT scans done earlier were described as normal, a very small granulomatous lesion enhanced by the contrast medium was found retrospectively in the last examination.

In addition to praziquantel and the anti-epileptic drug clonazepam, the patient received cimetidine to increase the drug's bioavailability[7–9] and a glucocorticoid to reduce the inflammatory reaction,[10,11] prednisone being preferred to dexamethasone as the latter may reduce the bioavailability of praziquantel.[7,12,13] This treatment regimen was well tolerated although seizures did significantly increase in number because of the inflammatory reaction caused by the death of the cysticerci. Serial MRIs showed progressive regression of the cerebral cystic lesions both in number and size with their total disappearance by September 1996. During the treatment period and after it, ELISA showed increasing antibody titers. EITB was positive for the specific 26 K and 8 K bands before treatment, weakly positive 3 months after treatment and negative for the 8 K band upon discharge in October 1996.

During the incubation period between infection with cysticerci and the development of symptoms which varies from less than 1 year to 30 years with an average period of about 5 years, the worm may have been expelled in the stools, as was the case with our patient, and found in only 15% of people with neurocysticercosis.[14] The average cyst measures only a few millimetres, but larger cysts measuring a few centimetres have also been described.[15,16] As a consequence of the fixation of the cysticercus larva in the brain, only small disturbances may be present during its life cycle. Generalised seizures are the common presentation of the parenchymal type and represents up to half of late-onset cases of epilepsy in endemic countries.[17] However, any neurological syndrome attributable to single or multiple small cerebral space occupying lesions can occur including mental disorders, extra-pyramidal disorders, intracranial hypertension, transient paresis, hemiparesis, meningoencephalitis, seizures and focal nervous lesions of the brain or the spinal cord. Ischaemic cerebrovascular disease, whether transient or irreversible, is an under-recognised and relatively common complication.[7,18–25] A fatal outcome is uncommon, usually being due to status epilepticus or the development of a life-threatening intracranial hypertension.[26]

Three different types of neurocysticercosis locations, cisternal, intraventricular and parenchymal, have been recognised and can be found simultaneously in the same patient reflecting different infectious episodes. Our patient harboured the parenchymal type which is found in more than two thirds of all known cases and in which four stages of the disease are recognised.[27,28] In the early vesicular stage, the cysticercus consists of a thin capsule surrounding a viable larva. The pathognomonic finding on MRI is a rounded CSF-like cyst with a mural nodule representing the scolex. Oedema and ring-like contrast enhancement are rare. In the second colloidal vesicular stage the larvum dies and releases metabolic products which generate host inflammatory response. Gadolinum injection reveals ring-like enhancement due to oedema. In the third granular stage the cyst retracts, its contents begin to mineralise and its capsule is quite thick. The lesion appears as an enhancing nodule with or without surrounding oedema similar in appearance to other granulomata.[29,30] The final stage is commonly found when no treatment has been given. The lesion is better seen on CT studies and appears as a small calcification without oedema or contrast enhancement. In our case, MRI showed the first three stages and was more sensitive than CT.

As a consequence, we postulate that the symptoms presented by this quadriplegic patient resulted from autonomic dysfunction and transient cerebral ischaemia secondary to the many space occupying cysts with surrounding inflammatory reaction and reactive vasculitis. Whether neurocysticercosis played a role in initiating AD is not totally clear at this point. However, once the treatment of neurocysticercosis was achieved, the patient was symptom-free. This might also have resulted from the prompt treatment of the various noxious stimuli and from the spontaneous temporal decay of AD which reaches a peak after a lapse of time following SCI and then subsides.[2] From a retrospective point of view, the early confusional states and transient aphasia episodes were compatible

with ictal and post-ictal states. Considering the small size of the intracerebral cysts, we might also postulate that smaller CT sections would have probably been able to reveal at least one of the cysts thus increasing the sensitivity of this particular examination. The occurrence of SCI in a patient harbouring cysticerci seems to be a pure coincidence. Whether or not the administration of high-doses of steroids in the immediate post-traumatic period has played a role in activating cysticercosis is not clear.

Conclusion

In a quadriplegic patient, the diagnosis of neurocysticercosis might be difficult because some of its symptoms may mimic those of autonomic dysreflexia and orthostatic hypotension which are frequent complications of high level SCI. Neurocysticercosis should be suspected when seizures and/or focal neurologic signs accompany AD in an otherwise healthy SCI patient living in or coming from an endemic zone. MRI is more sensitive than CT in the early diagnosis of active neurocysticercosis and thin contiguous sections should be preferred when the patient is investigated with both methods. Treatment of cerebral cysticercosis with praziquantel associated with cimetidine and prednisone gives good results. A recent search of the Medline database throughout the period 1966–November 1997 did provide a total of 1590 papers dealing with neurocysticercosis. Cases of medullary compression or local inflammatory reactions secondary to spinal cysticercosis have been reported. However, to the best of our knowledge, no case of cerebral neurocysticercosis accompanied by AD in a quadriplegic patient manifesting itself by AD has been described previously.

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