Rare disease

Psychiatric manifestations of neurocysticercosis in paediatric patients

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Summary

Neurocysticercosis is the most frequent parasitic disease of the central nervous system and is caused by the larval stage of the tapeworm *Taenia solium*, in which the human being becomes the parasite's intermediary host. The disease has a worldwide distribution, with a marked prevalence in underdeveloped or developing. However, this disease is rare in children, which is attributed to the long incubation period of the disease, ranging from several months up to 30 years. For this reason, very few cases are found in infants at the age of preschoolers, while most frequently cases are found in children between the ages of 6 and 16.

BACKGROUND

The aim of this paper is to review the literature about the different neuropsychiatric manifestations that have been observed in patients with neurocysticercosis. Likewise, particular attention will be paid to the distinctive features regarding the clinical expression of such manifestations in both infants and adolescents. The importance we attribute to such symptoms is the possibility of real debut with manifestations of this disease that are rarely related to this diagnosis (cognitive impairment, depression and psychosis), highlighting the importance of updating the need in order to consider it, given the increasing prevalence of these parasites due to immigration from endemic countries.

CASE PRESENTATION

Ecuadorian origin male patient, at the present time he is 15 years old. Two years ago the patient came to the emergency unit after suffering two generalised seizure episodes. According to what his mother said, the boy complained of headaches and dizziness since the age of 7. Between the ages of 7 and 13 he lived in Ecuador with the mother's family, without receiving medical care. Upon arrival to Spain, some 8 months prior to suffering the crisis, the patient is assessed by their primary care physician in relation to the above described symptoms, and is diagnosed with frontotemporal throbbing headache without photophobia, nausea or vomiting. The patient is prescribed to follow anti-inflammatory analgesic treatment. Given the lack of response to treatment, and after conducting basic analyses in which all values are within normality, the patient is then derived to a psychologist who in turn identifies his symptoms as a calling for attention behaviour due to the difficulties that convey in the adapting to a new life situation. At the moment of his visit to the emergency unit, the patient requires admission to pediatric intensive care unit (ICU) for 24 h. Blood tests with blood count, biochemistry and venous blood gases are conducted, all giving values within normal limits, as well as the determination of

toxic substances in urine and serum ethanol, where they are all negative. Cranial CT was performed in which suggestive findings of neurocysticercosis with multiple cortical, subcortical and subependymal damages at different stages of evolution were found. While at the ICU, the patient is administered valproic acid treatment and dexamethasone. The patient progresses well, without further convulsive episodes and maintains a good level of awareness. On the following day, the patient is taken to the paediatric floor with a suspicion of a possible neurocysticercosis diagnosis. An extended study for such parasitosis is then conducted: cysticercus serology with absence of antibodies, fecal parasites where *Blastocystis hominis* and *Entamoeba coli* are isolated, resulting negative for cryptosporidium. Cerebral MRI was performed with contrast (figures 1 and 2) which is reported as follows: Neurocysticercosis preferably with supratentorial parenchymal involvement, subarachnoid and intraventricular with over 100 damages, mostly in vesicular state, several in-vesicular colloidal stages and some calcified. Neurocysticercosis diagnosis was confirmed with secondary headaches and seizures. Later on, during his admission, a psychiatry-liaison takes place since the patient was sad, apathetic, showed concern for his somatic situation and had difficulties in concealing sleep. A diagnosis for adjustment disorder with depressive symptomatology is formulated, the patient is treated with antidepressants and anxiolytics and is then derived for monitoring to child psychiatry. When the patient is discharged, the treatment is the following: 500 mg of valproic acid every 8 h, 400 mg of Albendazole every 12 h, 10 mg of Fluoxetine in the morning and 5 mg of clorazepate dipotassium before going to bed, ending the tapering of corticosteroids on the day of discharge.

Three months after formulating the diagnosis, brain MRI without contrast control is performed, in which the evolution of practically all injuries to a nodular granulomatous / calcified stage are observed. Periodic electroencefalographic records are also conducted for control, observing in all

Figure 1 Multiple cortical, subcortical and subependymal damages at different stages of evolution were observed in the cranial CT.

cases, normal activity without focal or associated generalised epileptiform abnormalities.

From the moment of the patient's admission up to the present day, the patient has had in numerous occasions to attend the emergency unit due to seizures, in principle due to by the irregular administration of the anticonvulsant medication and further on due to their poor handling that has required some adjustment of the treatment. During this period, the patient follows regular Neuropediatrics, infectious diseases and child psychiatry monitoring.

Since the first review in child psychiatry, the patient expresses fear to leave home alone, fear to fall in the street, does not feel like doing anything, and as a result spends most of the day watching television or in bed because his head hurts. There is no loss of appetite or weight, but has difficulty falling asleep, frequent awakenings besides those due to his headaches. His mother states that he looks down, inexpressive and that before the seizure, he was already different, with great irritability and alterations in his behaviour. The diagnosis formulated during admission is confirmed as adjustment disorder with depressive symptomatology and the dose of fluoxetine is increased to 20 mg per day. During the following months, clinical stability is achieved in regard to headaches, seizures and mood, reporting however, a significant deterioration in his behaviour: he is disobedient, defiant and constantly fights with his mother and brother. It was decided upon psychological intervention for the patient's adaptation problem and dysfunctional family dynamics. After a further seizure, somatic concerns reappear significantly, increasing irritability and oppositional attitude, challenging and capricious behaviour, as well as the worsening of insomnia conciliation. 1 mg of Lormetacepam treatment is added at bedtime. In the following months the patient

develops an even lower impulsivity control, becoming at times very demanding and even aggressive, alternating episodes in which he is very withdrawn, quiet, with others of great aggressiveness and irritability. The patient himself is aware of his difficulties in concentration, affecting academic performance.

DISCUSSION

The study of mental disorders in patients with cysticercosis has a very important historical origin. It is so, that the first clinical disease classification was in 1862 developed by the German neuropsychiatrist Griseinger.^{1,2} This classification was made based on psychiatric symptoms in these patients, classifying them in four groups: epileptic patients without mental illness, epilepsy patients with mental illness, patients with chronic mental illness alone, and patients with signs and symptoms of diffuse cerebral irritation. On this basis, for years, many patients with neurocysticercosis were treated in psychiatric institutions, without a correct etiological diagnosis until the onset of epilepsy or other neurological signs raised the suspicion of parasitic disease.^{3,4}

The production mechanisms of mental disorders in this disease have not been widely studied for the moment. If it is clear that the presence of parasites can cause structural changes in the central nervous system (CNS), with significant involvement of immune mechanisms, leading to oedema, reactive gliosis and arachnoiditis, which in turn can influence proliferative endarteritis with occlusion of the lumen, and hydrocephalus, and as a result of all this will lead to intracranial hypertension and cerebral oedema.^{1,5-9} These mechanisms, in turn, depend upon the parasite burden (number and size) and location of parasites in strategic areas of a cognitive and an affective level. It is

Figure 2 Cerebral MRI was reported as follows: neurocysticercosis preferably with supratentorial parenchymal involvement, subarachnoid and intraventricular with over 100 damages, mostly in vesicular state, several in-vesicular colloidal stages and some calcified.

also important to keep in mind the importance of psychosocial factors surrounding the disease, and in particular the discomfort of headaches, seizures, and loss of functionality in a significant proportion of patients.

In general, clinical features are similar in children and adults, although there are some significant differences that will be described in detailing.^{2–4} The clinical picture is characterised by episodes of exacerbation and remission, depending upon the evolutionary time in which the patient is assessed, as well as in the number, the size and the location of the cysts in the CNS. The stages of deterioration occur because of death and disintegration of the parasite and the subsequent intensification of the inflammatory reaction described above. Cysticercoids may remain asymptomatic for years if they are located in brain areas lacking eloquence and with the existence of immunological tolerance. In many cases, children maintain the disease in an asymptomatic form for some time.^{4 10–13}

The current symptom classifications groups symptoms according to most frequent and constant manifestations:

1. **Convulsive form:** The most common appearance is the occurrence of epileptic seizures in both adults and children, being most frequent in the latter. In adulthood they may be the only initial manifestations of the disease, appearing most frequently in the form of quísticas cortico-meníngeas and only in 15% of those on basal locations. Complex partial seizures and secondarily generalised are more frequent, tonic-clonic seizures being more frequent in second place. Moreover, in children, the possibility of seizure recurrence is greatest. A motor deficit may also appear, being less frequent in children.^{1 2 12}
2. **Hypertensive form:** Intracranial hypertension syndrome is also very frequent, usually with expression in

subacute or chronic forms (holocránea headache, projectile vomiting and papilloedema) and, less commonly, acute (intense headache with loss of consciousness). A clinical association may appear, being characteristic of a focal deficit, cognitive impairment and seizures. It is less frequent in children than in adults, there can be various cysts, mild to moderate oedema may be generated and a relatively benign and significant progressive course. At times sudden episodes of faintness and headaches, specially that of flexo-extension (Bruns' Syndrome) may appear, due to the presence of a cyst in the IV ventricle.^{1 2 12}

Form insane: Neurocysticercosis progressively alters brain function, producing a progressive organic brain damage (usually begins with changes in memory and computation). Mental alterations are the initial symptom in the 2.3% of patients.² It normally begins with alterations in memory, ability to calculate and in the capacity for the abstraction. Sometime in the development the following appear: alterations in language apraxia de la marcha, memory alterations and cognitive impairment. This symptomatology is often associated with psychiatric manifestations (irritability, delusions, hallucinations, psychomotor excitement, apathy, depression, disinhibition of instincts, personality disturbance, social maladjustment...). In its chronic form, clinical pictures are presented with compatible characteristics with the demential, maniforme or squizoid syndrome. While in adults predominates neurocognitive deficits and psychiatric morbidity in children predominates more significant difficulties in learning and behavioral changes.^{1 2 12}

The patients of our case report presents, as it has already been mentioned, more than 100 damages located in different areas of the brain parenchyma, as well as in ventricular

spaces as can be seen in the attached figures. The observed behavioural alterations may be explained due to damages in the frontal lobes, even though it will be difficult to correlate each symptom with a specific anatomical area.

As to the evolution and forecast of the disease, we are dealing with a dangerous and aggressive disease which develops progressively to an accentuated intellectual impairment. A high seizure recurrence is frequent in children.

No scales have been used to measure psychopathology, only clinical assessment has been conducted for the described symptoms.

Learning points

There are some other differences in childhood neurocysticercosis regard to adult:

- ▶ In many cases, children remain asymptomatic disease for long periods of time.
- ▶ Have almost exclusively parenchymal different forms, with subarachnoid and intraventricular locations rare. Cysticerci encephalitis is more common in this age.
- ▶ The injury most commonly found in childhood is the cystic degeneration, or transitional, usually single, which debuted with symptomatic seizures

Competing interests None.

Patient consent Obtained.

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