Case Report

Hypothalamic neurocysticercosis presenting with polyuria: a first report of an unusual manifestation

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Introduction

Neurocysticercosis is the most common cystic lesion to occur in the central nervous system, especially in developing and tropical countries. Dissemination of infection with cysticercus occurs in humans after the ingestion of the eggs of Taenia solium in contaminated food (accidental intermediate host), after the regurgitation of eggs, from the intestine into the stomach, in a patient who harbours the adult worm (internal auto-infection), or carried by unclean hands (autoinfection). Larvae or oncospheres invade the gastrointestinal mucosa and spread haematogenously to other organs. The most commonly affected organs are the brain, eyes and skeletal muscles. The characteristic lesion of cysticercus is a cyst with an internal scolex. Although 80% of the cases of neurocysticercosis are asymptomatic, patients may present with seizure, headache, symptoms of hydrocephalus or increased intracranial pressure, visual blurring due to papilloedema, visual field defect and diplopia. Hormonal disturbances are rare presentations. Neurocysticercosis of the pituitary gland and hypothalamus are so uncommon that only a few cases have been reported in the literature [1–4]. We report a patient with neurocysticercosis who presented with polyuria. Tests revealed complete central diabetes insipidus (DI) and panhypopituitarism resulting from hypothalamic and pituitary gland involvement by neurocysticercosis.

Case

An 81-year-old Thai male was admitted to our hospital with the chief complaint of increase in the frequency and amount of urination for 1 month. The patient had been well until 3 months before admission, when he developed increased urinary frequency (5 times during the day and 5 times overnight). He also had increased thirst and water intake (3 l and 1.5 l during the day and overnight, respectively) and poor appetite and constipation. His daily activities were impeded. One month before admission, he had polyuria, up to 10 times during the day and 10 times at night, for which he visited a clinic. He had a normal fasting blood sugar (6.1 mmol/l). A computed tomography (CT) of his brain showed multiple parenchymal brain calcifications. The patient was therefore referred to our department. He revealed a poor appetite and a weight loss of 6.6 pounds over 3 months. He denied having hesitation during urination or dribbling after urination. He did not have a history of fever, headache, myalgia, visual blurring, double vision, diabetes mellitus, hypertension, coronary artery disease or seizure disorder.

His temperature was 36.0°C, respiration rate 16 per minute, blood pressure 120/80 mmHg, and pulse 90 beats per minute. He did not have a puffy face and eyelids or enlarged thyroid gland. His axillary and pubic hair were normal in appearance for his age. His neurological examination was normal, with no visual field defect. The rest of his examination was unremarkable.

Initial laboratory tests showed the following: haemoglobin 13.3 g/dl, white cell count 10.2 × 109/μl (10.2 × 109/l), platelet count 397 × 109/μl (397 × 109/l), serum sodium 140.6 mmol/l, potassium 4.2 mmol/l, chloride 101 mmol/l, carbon dioxide 28.6 mmol/l, urea 2.6 mmol/l, creatinine 89 μmol/l, calcium 2.44 mmol/l, albumin 41 g/l, normal liver function tests. His urinalysis showed a pH of 6.0 and a specific gravity of 1.000, with no protein, sugar, or white or red cells or casts.

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His chest X-ray and electrocardiogram were normal. His serum and urine osmolalities were 302 and 85 mosmol/kg, respectively. Urine electrolyte levels were: sodium 11.7 mmol/l, potassium 6.26 mmol/l, chloride 11.4 mmol/l.

His initial presumptive diagnosis was water diuresis probably due to DI. The patient underwent a water deprivation test. At the beginning of the test, after water deprivation, his laboratory test results were: serum sodium 144.3 mmol/l, potassium 3.4 mmol/l, chloride 106.3 mmol/l, carbon dioxide 28.7 mmol/l, serum osmolality 307 mosmol/kg, urine osmolality 59 mosmol/kg and urine specific gravity 1.000. After DDAVP was given, his urine osmolalities at 30, 60 and 120 min were 69, 332 and 384 mosmol/kg, respectively.

Magnetic resonance imaging (MRI) without enhancement, of his brain, revealed multiple tiny lesions in both cerebral hemispheres, and cerebellum, hypothalamus, infundibulum and pituitary gland (Figures 1 and 2). The results of the water deprivation followed by the DDAVP test and the imaging study suggested DI secondary to cysticercosis, which was confirmed by a positive enzyme-linked immunoelectrotransfer blot (EITB) test for cysticercosis. Because the lesions were found in the hypothalamus, infundibulum and pituitary gland, pituitary function tests were done with the following results:

<table>
<thead>
<tr>
<th>Test</th>
<th>Result</th>
<th>Reference Range</th>
</tr>
</thead>
<tbody>
<tr>
<td>Serum cortisol (08.00 a.m.)</td>
<td>1.13 µg/dl</td>
<td>(5–25)</td>
</tr>
<tr>
<td>Repeated serum cortisol</td>
<td>0.97 µg/dl</td>
<td></td>
</tr>
<tr>
<td>Free T4</td>
<td>0.426 ng/dl</td>
<td>(0.932–1.71)</td>
</tr>
<tr>
<td>Thyroid-stimulating hormone (TSH)</td>
<td>1.71 mU/l</td>
<td>(0.27–4.20)</td>
</tr>
<tr>
<td>Follicle-stimulating hormone (FSH)</td>
<td>60.57 ng/ml</td>
<td>(4.1–18.4)</td>
</tr>
<tr>
<td>Testosterone</td>
<td>&lt;0.02 ng/ml</td>
<td>(3–10)</td>
</tr>
</tbody>
</table>

Panhypopituitarism was diagnosed, because the patient had central hypothyroidism, secondary or tertiary adrenal insufficiency and low FSH at the same time as low testosterone. He was treated with prednisolone 5 mg every morning and 2.5 mg every evening, levothyroxine 50 µg per day orally and DDAVP 10 µg per day intranasally. Urinary frequency completely resolved and he returned to normal life and daily activities.

**Discussion**

This patient presented with polyuria, which could be caused by many conditions, such as DI, primary polydipsia or solute diuresis. His initial very low urine osmolality excluded the possibility of solute diuresis as the cause of polyuria. The next step in his evaluation was a standard dehydration test, in which the urine osmolality of 59 mosmol/kg after water deprivation confirmed the diagnosis of DI. Central DI was the most likely diagnosis because the patient was found to have multiple lesions in the brain (including the hypothalamus, infundibulum and pituitary gland), and pituitary function tests revealed panhypopituitarism. The differential diagnosis of the brain lesions included cysticercosis, echinococcosis, tuberculoma and toxoplasmosis. Echinococcosis usually forms a large hydatid cyst along with daughter cysts with perilesional inflammation. In toxoplasmosis and tuberculoma, brain lesions are usually ring-enhancing lesions and abscesses are more common than cysts. Toxoplasmosis is
commonly found in an immunocompromised host, and is accompanied with progressive clinical deterioration.

The definitive diagnosis of cysticercosis was made based on two major criteria [5]. The characteristic lesions seen on the MRI of our patient suggested neurocysticercosis, and a positive serum EITB assay for cysticercosis confirmed the diagnosis. The current literature indicates that the enzyme-linked immunoelectrotransfer blot (EITB) assay for cysticercosis has a sensitivity of 86–98% and a specificity of 92.8–100% [6]. In addition, in developing and tropical countries, neurocysticercosis is the most common cystic lesion in the central nervous system. Interestingly, the MRI detected the hypothalamic lesion which went along with the high serum prolactin. It is known that the prolactin inhibitory factor is secreted in the hypothalamus and it travels along the pituitary stalk to the pituitary. So, a high prolactin level would be expected if there are lesions in the hypothalamus or pituitary stalk.

Infestations of the pituitary gland and hypothalamus by cysticercosis are uncommon, and only a few such cases have been reported. Hormonal disturbances have been rare presentations. Only two cases of panhypopituitarism [3,4], two cases of galactorrhea [3] and one case of diabetes insipidus (leading to death) [2] caused by cysticercosis were reported. On postmortem examination, two patients were reported to have cysticercus in the ventromedial nucleus of the hypothalamus – associated with obesity [1]. However, a patient presenting with polyuria resulting from the neurocysticercosis of the hypothalamus has not been reported before. In our patient, because all lesions seen on MRI were calcified, and there was neither perifocal oedema nor abnormal enhancement in these spots, there was no need to treat the nonviable (dead) cysticerci [7].

The polyuria seen in our case was the result of complete central DI associated with neurocysticercosis, a polyuria of unusual etiology related to a tropical disease.

Conflict of interest statement. None declared.

References
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