Bruns Syndrome Caused by Intraventricular Neurocysticercosis: Case Report and Literature Review

Síndrome de Bruns causada por neurocisticercose intraventricular: Relato de caso e Revisão da literatura

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Introduction

Neurocysticercosis is the most common helminth infection of the central nervous system, caused by the larval form of Taenia solium. Bruns syndrome, described in 1906 by German neurologist Ludwing Bruns, is one of the presentations of intraventricular neurocysticercosis. It is characterized by a transient increase in intracranial pressure, caused by a floating mass in the ventricular system, producing hydrocephalus and leading to episodes of headache, vertigo and vomiting, which are triggered by sudden head movements.1,2 The authors report the case of a patient who presented Bruns syndrome due to neurocysticercosis. A review of the published cases is discussed in the present article.

Case Presentation

The patient was a 46-year-old female, from the countryside of the state of Rio Grande do Sul, Brazil, with a history of untreated hydrocephalus, cognitive deficit and bacterial meningitis treated in the previous two years. Over the course of two weeks, she presented recurrent episodes of syncope, paresis in the four limbs, and loss of sphincter control, which improved between crises, and was associated with progressive visual loss, without improvement since the onset of the condition. Four days after the onset of symptoms, she had an episode of seizure and vomiting. With the use of phenobarbital and phenytoin, she managed to control the seizures.

Keywords

► neurocysticercosis
► cysticercus
► neurosurgery

Abstract

Bruns syndrome is one of the clinical presentations of intraventricular neurocysticercosis, and it is characterized by episodes of headache, vertigo and vomiting. The intraventricular form of neurocysticercosis occurs in 7% to 20% of the cases, and it is more serious than the intraparenchymal form. The management is primarily surgical, associated with pharmacological therapy with anthelmintic drugs and corticosteroids. We report the case of a patient who presented Bruns syndrome due to neurocysticercosis.

Palavras-chave

► neurocisticercose
► cisticero
► neurocirurgia

Resumo

A síndrome de Bruns é uma das apresentações clínicas da neurocisticercose intraventricular, caracterizada por episódios de cefaleia, vertigem e vômitos. A forma intraventricular da neurocisticercose ocorre entre 7% e 20% dos casos, e é mais grave do que a forma intraparenquimatosa. O tratamento é principalmente cirúrgico, associado a terapia farmacológica com drogas anti-helmínticas e corticoides. Relatamos o caso de uma paciente que apresentou síndrome de Bruns por neurocisticercose.

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The patient started an investigation with a neurologist at Santa Casa de Misericórdia de Porto Alegre, who requested a magnetic resonance imaging (MRI) scan. She sought the emergency department of the same institution after experiencing worsening of the paresis in the limbs and difficulty in walking. In the MRI scan, two cystic lobulated and septate lesions within the fourth ventricle were identified, causing ventricular expansion, with obstruction of the cerebral aqueduct, associated with an important dilation of the supratentorial ventricular system. Cystic lesions were also identified in the topography of the cisterna magna (►Figs. 1 and 2).

Upon physical examination, the patient was confused. Ocular opening was spontaneous, and the eyes obeyed bilateral commands.

A hypothesis of intraventricular racemic neurocysticercosis as the cause of Bruns syndrome was considered. After discussion with the neurosurgery team, the patient was admitted to Hospital São José and referred for surgical removal of the cysts. In the immediate postoperative period, the patient persisted with reduced level of consciousness and random eye movements. The postoperative tomography showed a diffuse reduction in the amplitude of the cortical sulci and gyrus, associated to loss of cortical-subcortical differentiation and reduction in the size of the basal cistern, an aspect related to diffuse cerebral edema. The ventricular system was effaced, and pneumoventricle in the frontal horns of the lateral ventricles was observed (►Fig. 3), which required the placement of and external ventricular shunt, and, subsequently, a ventricular-peritoneal shunt (VPS). After a positive Weinberg test, drug therapy with albendazole and dexamethasone was initiated. The patient developed into a persistent vegetative state and died 46 days later due to respiratory failure.

Discussion

The clinical presentation of neurocysticercosis may vary depending on the host's immune status, the size of the cyst, and its location in the central nervous system. The
most common symptoms are epilepsy, headache, and focal neurological deficit, and the intraventricular form of neurocysticercosis only occurs in 7% to 20% of the cases, with the third and fourth ventricle being the most affected sites.\textsuperscript{1,3}

The main causes of Bruns syndrome are mobile intraventricular lesions such as tumors, neurocysticercosis and colloid cyst of the third ventricle, with neurocysticercosis being the most common cause.\textsuperscript{4} The syndrome consists of episodes of intense headache, vomiting and vertigo triggered by sudden movements of the head that produce transient hydrocephalus by displacement of the mobile ventricular mass. Some cases may present with rapid neurological worsening and death due to intracranial hypertension.\textsuperscript{5} The diagnosis is difficult because the clinical manifestations are not very specific. The MRI is superior to tomography for the diagnosis of neurocysticercosis.\textsuperscript{5}

When the ventricular form is accompanied by meningitis and hydrocephalus, mortality is greater than 50%, and most patients die within 2 years, even with the VPS. The prognosis of the intraventricular form is worse than that of the intraparenchymal form, and it follows a progressive course, due to obstructive hydrocephalus, intracranial hypertension and meningeal infection.\textsuperscript{5} The clinical presentation of our case was less common because the patient only had visual loss on the neurological examination, despite signs of hydrocephalus and occlusion of the fourth ventricle in the MRI.

The management of intraventricular neurocysticercosis is different from that of the intraparenchymal form. Surgery is the primary therapeutic measure for the removal of the cysts. For cysts of the third and lateral ventricles, the endoscopic approach is the most recommended. In cases of involvement of the fourth ventricle, removal of the cysts may be more challenging, requiring open surgery. Ventricular shunts are commonly required for the management of obstructive hydrocephalus. Our patient underwent a similar management to that reported in the literature, with surgical removal of the cysts and associated drug therapy. The telovelar approach was chosen for the removal of the cysts

\begin{table}
\centering
\begin{tabular}{|l|l|l|l|l|l|}
\hline
Author & Age/Gender & Location & Clinical presentation & Management & Outcome \\
\hline
Aguilar-Amat et al\textsuperscript{1} & 29y/F & Intraventricular\textsuperscript{**} & Headache, nausea, diplopia and gait instability & Albendazole, corticoids and surgery\textsuperscript{7} & No deficits \\
\hline
Rodriguez et al\textsuperscript{8} & 43y/F & Fourth ventricle & Headache, vomiting and vertigo & Open surgery & – \\
\hline
Jensen and Post\textsuperscript{7} & 50y/F & Foramen of Monro & Headache, nausea, irritability and confusion & Endoscopic surgery & No deficits \\
\hline
Jensen and Post\textsuperscript{7} & 30y/F & Foramen of Monro & Headache, fever and confusion & Endoscopic surgery & – \\
\hline
Roongpiboonsopit et al\textsuperscript{4} & 69y/M & Fourth ventricle & Headache, vomiting and confusion & Surgery\textsuperscript{*} & No deficits \\
\hline
Torres-Corzo et al\textsuperscript{3} & 22y/M & Third ventricle & Headache and vomiting & Endoscopic surgery & No deficits \\
\hline
Torres-Corzo et al\textsuperscript{3} & 31y/F & Third ventricle & Headache and papilledema & Endoscopic surgery & No deficits \\
\hline
Torres-Corzo et al\textsuperscript{3} & 17y/F & Third ventricle & Headache and visual alterations & Endoscopic surgery & No deficits \\
\hline
Torres-Corzo et al\textsuperscript{3} & 28y/M & Third ventricle & Headache and vomiting & Endoscopic surgery & No deficits \\
\hline
Torres-Corzo et al\textsuperscript{3} & 43y/M & Third ventricle & Lethargy & Endoscopic surgery & No deficits \\
\hline
Torres-Corzo et al\textsuperscript{3} & 54y/M & Third ventricle & Headache, vomiting and diplopia & Endoscopic surgery & No deficits \\
\hline
Torres-Corzo et al\textsuperscript{3} & 35y/F & Third ventricle & Headache and Parinaud sign & Endoscopic surgery & No deficits \\
\hline
Shahani et al\textsuperscript{2} & 40y/F & Fourth ventricle & Headache, nausea and vomiting & Albendazole, dexamethasone, surgery\textsuperscript{*} and VPS & No deficits \\
\hline
Shahani et al\textsuperscript{2} & 39y/F & Fourth ventricle & Headache, dizziness, nausea, vomiting and tinnitus & Albendazole, dexamethasone, surgery\textsuperscript{*} and VPS & No deficits \\
\hline
Das et al\textsuperscript{6} & 24y/M & Fourth ventricle & Headache, vertigo and vomiting & Surgery\textsuperscript{*} & No deficits \\
\hline
Dhiman et al\textsuperscript{9} & 11y/M & Fourth ventricle & Headache, vomiting and strabismus & Endoscopic surgery & Maintained strabismus \\
\hline
\end{tabular}
\caption{Review of published cases of intraventricular neurocysticercosis}
\end{table}

Abbreviations: F, female; M, male; VPS, ventriculoperitoneal shunt; Y, years old.
Notes: *Open or endoscopic surgery not specified. **No report of which ventricle was affected.
from the fourth ventricle. The flexible endoscopy approach is an option for patients with neurocysticercosis in the third ventricle. The treatment with anthelmintic drugs may lead to worsening of the symptoms, and it should be administered concomitantly with glucocorticoids. The anthelmintic drug of choice is albendazole, which is superior to praziquantel because it does not interact pharmacologically with glucocorticoids and antiepileptic drugs.

In a review of the PubMed database with the MeSH terms neurocysticercosis AND Bruns syndrome, eight publications of Bruns syndrome caused by neurocysticercosis were found, totaling 16 cases. Headache was the main symptom reported (15 cases), followed by vomiting (9), confusion (3) and vertigo (3). Visual and ocular motility disorders occurred in six cases. Gait instability, tinnitus and fever were reported in one case each. The age of the patients ranged from 11 to 69 years, and surgical intervention was the choice in all cases. In the present case, syncope, paresis and visual loss were the initial symptoms. Signs of intracranial hypertension, such as vomiting, appeared a few days later. The review of the published cases is summarized in Table 1. The involvement of the third and fourth ventricles was similar, with 7 and 6 cases respectively, and in 2 cases the cysticercus was located in the lateral ventricles, in the foramen of Monro. Endoscopic surgery was the choice in most cases. The patient maintained the neurological deficit only in one case.

Conclusion
Bruns syndrome is a rare presentation of neurocysticercosis. It has a nonspecific symptomatology that can go unnoticed by the physician, becoming a serious condition and leading to death due to obstruction of the flow of the cerebrospinal fluid.

Conflict of Interest
The authors have none to disclose.

References