v. 2, n. 29, 2022

International Journal of Health Science

HYDROCEPHALIA DUE TO RACEMOSA NEUROCYSTICERCOSIS: REPORT OF TWO CASES

Julia Brasileiro de Faria Cavalcante
Neurosurgery Resident Physician at the institution: Hospital de Neurologia Santa Mônica, Goiânia, Goiás – Brazil
http://lattes.cnpq.br/8529681469406105

Pedro Nogarotto Cembraneli
Neurosurgery Resident Physician at the institution: Hospital de Neurologia Santa Mônica, Goiânia, Goiás – Brazil
http://lattes.cnpq.br/6881953147326054

Ítalo Nogarotto Cembraneli
Graduating from the Medical Course of the institution: Centro Universitário de Mineiros (UNIFIMES), Mineiros, Goiás – Brazil
http://lattes.cnpq.br/9802814450132395

Renata Brasileiro de Faria Cavalcante
Preceptor of the Medical Residency at Hospital de Neurologia Santa Monica, Member of the Brazilian Society of Neurosurgery, Goiânia, Goiás – Brazil
http://lattes.cnpq.br/4940570467357094

José Edison da Silva Cavalcante
Professor, Doctor, PhD, Member of the Brazilian Society of Neurosurgery, Head of the Neurosurgery Residency at the institution: Hospital de Neurologia Santa Mônica, Goiânia, Goiás – Brazil
http://lattes.cnpq.br/8506840484334261

Abstract: Cysticercosis is a parasitic disease caused by the larval stage of Taenia solium (T solium), which when it reaches the Central Nervous System (CNS) generates neurocysticercosis. The clinical manifestations are varied, and can generate epileptic seizures, meningitis, hydrocephalus, among others. The diagnosis of neurocysticercosis is performed through neuroimaging (computed tomography and magnetic resonance imaging) and examination of cerebrospinal fluid (CSF). Currently, the first line of treatment is based on the use of anthelmintic associated with corticosteroids, and when necessary, neurosurgery.
Keywords: Neurocysticercosis, *Cysticercus racemosus*, Neurosurgery.

INTRODUCTION

Human cysticercosis is a parasitic disease caused by: *Taenia solium* (T. solium) *in the larval stage, considered by the World Health Organization (WHO) a public health problem, as well as a neglected infection, with predominance in countries such as: Latin America, Africa and Asia. (1-4).*

The cysticercus reaches the CNS and a cyst develops (*Cysticercus cellulosae*) which can lodge in different regions such as: brain parenchyma, subarachnoid space, meninges, medulla and ventricular system, where it takes on a shape with vesicles that unfold, forming a cluster (Cysticercus racemosus). The clinical picture depends on the location and its activity. (5-7).

CASE REPORT, NUMBER 1

Male patient, 72 years old, farmer, living in a rural area all his life, initially without basic sanitation, drinking water from an artesian well and bathing in a river. In the last three months, he presented holocranial headache, intermittent, pulsating, with progressive worsening of pain, currently with intensity 7/10 on the subjective pain scale, with partial improvement when using simple analgesic and non-steroidal anti-inflammatory drugs.

Associated with this, in recent weeks, he presented difficulty walking, urinary incontinence and mental confusion. Magnetic resonance imaging (MRI) of the skull was performed, which showed marked tetraventricular hydrocephalus, with cerebrospinal fluid transudation, thinning with superior displacement of the corpus callosum, as well as effacement of cortical sulci and fissures in the convexity. In addition, multiple thin-walled vesicular lesions were detected, involving the basal cisterns, especially the chiasmatic, prepontine and magna cisterns, which are associated with racemose neurocysticercosis. (Figure 1).

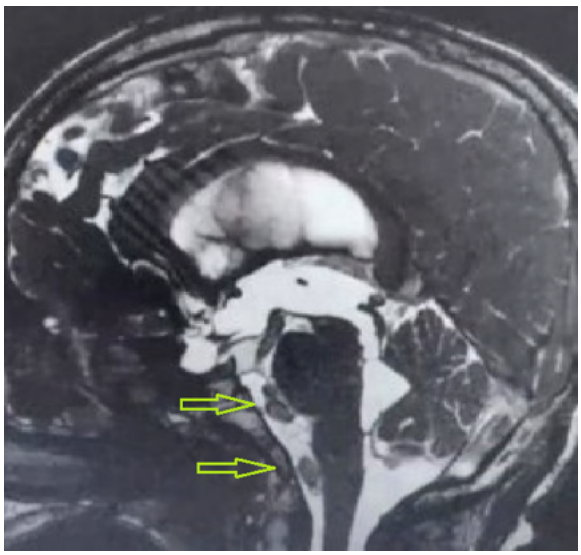

Figure 1. Sagittal MRI of the skull in Fiesta sequence showing thin-walled vesicular lesions associated with racemose neurocysticercosis.

Treatment with anthelmintic medication and corticosteroid therapy was started, without availability of serological test for neurocysticercosis. We opted for the surgical ventriculoperitoneal shunt, without intercurrences.

Control cranial computed tomography (CT) showed improvement in

hydrocephalus, and clinically the patient showed improvement in ambulation, urinary incontinence, improved level of consciousness and total improvement in headache. The patient was discharged from the hospital on the third postoperative day and continued outpatient follow-up.

CASE REPORT, NUMBER 2

A 58-year-old male patient, living in a rural area, presented sudden mental confusion associated with nystagmus, difficulty walking, severe headache, fever and convulsive crisis. Lumbar puncture was performed to study the CSF, showing a clear, colorless aspect, with opening pressure above the expected average, leukocytosis, glucose and protein consumption four times above the reference value.

Cranial CT showed significant dilatation of the supratentorial ventricular system, findings that suggest the possibility of CSF flow disorders. Laboratory tests showed leukocytosis and quantitative C-reactive protein above the reference value.

Cranial MRI was performed with a study of cerebrospinal fluid flow, showing multiple expansive cystic formations located in the cisterns of the base, the largest of which was located in the cisterna magna, measuring about 2.8x,1.8x2.2 cm in its major axes, determining compression on the floor of the fourth ventricle, compromising CSF flow, with consequent supratentorial and discrete infratentorial dilatation, with signs of CSF transudation. The set of alterations is compatible with an infectious/inflammatory process of the brain, racemose neurocysticercosis type. (Figure 2).

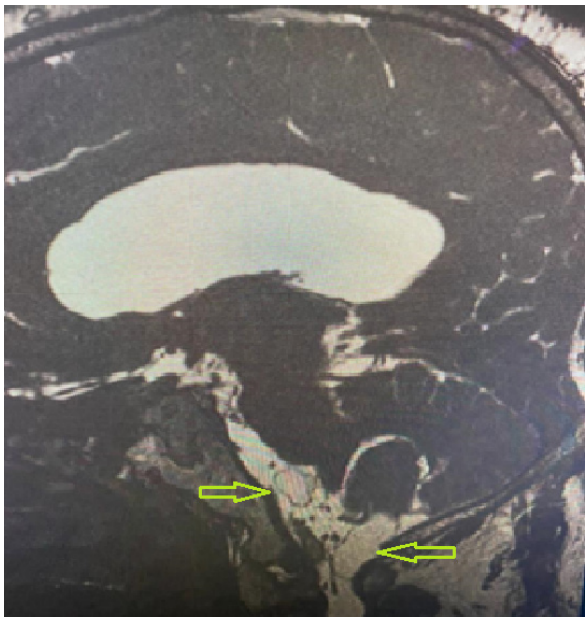

Figure 2. Sagittal MRI of the skull in the Fiesta sequence thin-walled vesicular lesions associated with racemose neurocysticercosis.

We opted for the approach to the lesion and surgical excision of the cysticerci (Figures 3 and 4), in addition to clinical treatment with corticosteroid therapy and anthelmintic medication.

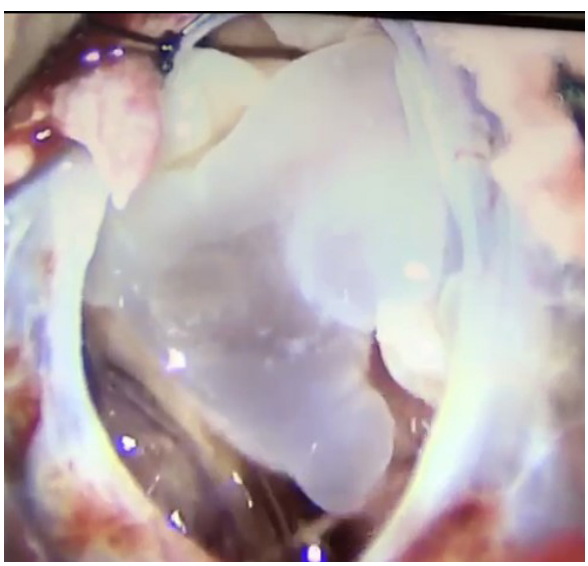

Figure 3. Intraoperative image showing racemose cysticerci filling the subarachnoid space.

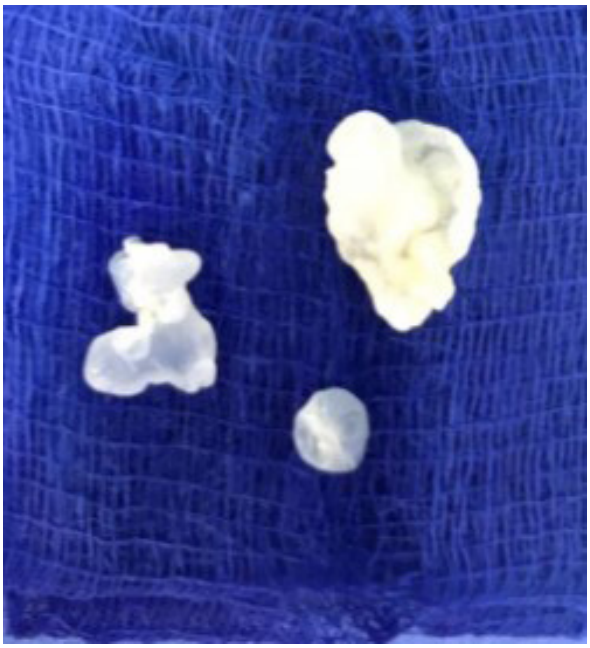

Figure 4. Racemose cysticerci removed during surgery. Appearance in grape bunches and absence of scolex.

The patient evolved on the second postoperative day with reduced level of consciousness, difficult-to-control convulsive crisis, severe headache and vomiting. Control cranial CT showed increased dilation of the ventricular cisterns. The neurosurgery team was called and performed an emergency external ventricular shunt (EVD).

On the fifth postoperative day, the EVD was removed, but the patient returned to present intracranial hypertension (ICH) symptoms. We opted for the surgical placement of a continuous lumbar drain with partial improvement of symptoms. Patient is in an Intensive Care Unit (ICU) bed for treatment of clinical complications resulting from prolonged hospitalization.

DISCUSSION

T. solium is a tapeworm that causes two distinct presentations in humans, depending on its life cycle: taeniasis and cysticercosis, affecting approximately 2.56-8.30 million individuals.(8-9)

Taeniasis refers to intestinal infection by adult tapeworms and occurs when infected undercooked pork is eaten. Although taeniasis is not associated with serious illness, humans shed T. solium eggs, which can infect pigs and other conspecifics. The result is the formation of cysts in the muscles, skin, eyes, or central nervous system. (5,10,11)

In humans, neurocysticercosis can result in severe disease, depending on the number, location, burden of cysts, and the resulting inflammatory host reaction. Most are asymptomatic, however, the most common clinical manifestation is a seizure. Neurocysticercosis is believed to be the leading cause of preventable epilepsy worldwide and can also cause chronic headache and hydrocephalus. (11.12)

The clinical symptoms of neurocysticercosis depend on the location, the number of cysts, the evolutionary stage of the lesions and the host's immune response. A single degenerated parasite is referred to as a cysticercus and may become calcified, appearing radiologically as a single nodular or cystic lesion with surrounding edema and contrast uptake. Cysticercus often resolves within a year of presentation, even without cysticide drug therapy. It is not known how many patients with cysticerci have seizures; however, a long-term follow-up study demonstrated an increased frequency of seizures in patients with associated perilesional gliosis, seen on MRI. (13-15)

Extraparenchymal neurocysticercosis is associated with hydrocephalus, meningitis, focal neurologic deficits, and death if not properly treated, usually requires neurosurgery (16)

Neuroimaging with CT and MRI is considered the gold standard for the diagnosis of neurocysticercosis. CT is sensitive for diagnosing intraparenchymal neurocysticercosis. MRI is sensitive for

recognizing parasites and visualizing scolex, parasitic degeneration, small cysts, subarachnoid cysts within the posterior fossa, spinal and basal cisterns, and cysts located within the ventricles, brainstem, cerebellum, and eye. (11,17,18)

Serological testing for neurocysticercosis in low-income countries is difficult, due to lack of testing, low sensitivity in patients with solitary or calcified cysticerci. Treatment with anthelmintics cannot be initiated without recent neuroimaging to exclude hydrocephalus, cysts at critical sites, and increased pressure, which are contraindications to anthelmintic drugs used alone. (19.20)

Anti-inflammatory therapy, such as corticosteroids, is commonly used to control inflammation. The current expert consensus is that anthelmintic drugs in combination with corticosteroids and anticonvulsants are beneficial in most patients with viable parenchymal cysts. (19-24)

Neurological symptoms such as epileptic seizures, headache, vertigo and vomiting are frequently reported during the first days of anthelmintic treatment, presumably due to the perilesional edema caused by the treatment; therefore, corticosteroids must be administered concomitantly. (22.24-26)

There are questions about the effectiveness of parasiticide drugs in the cisternal or intraventricular location and in the racemose form, recommending surgical extirpation as the best option. (27-29)

Ventricular involvement predominates in the fourth ventricle, where these lesions may be adhered to the ependymal plane or found free in the ventricular cavity, causing temporary obstruction of CSF flow, which leads to hydrocephalus, a condition that can also be caused by cysticercotic arachnoiditis. (2, 3, 30)

Surgical treatment is considered in cases where cystic lesions or basal arachnoiditis generate hydrocephalus in order to restore normal CSF flow, or in cases where the cystic lesion itself causes ICH. (31.32)

Neurocysticercosis is primarily a disease of poverty that predominantly affects rural populations with poor sanitation. Such pathology overloads the health system, in addition to generating an impact on the economy and society. The disease also affects swine farmers economically, as they may lose income if they cannot sell infected animals and meat. (33)

New cases can be prevented with community health and education interventions involving: vaccination and anthelmintic treatment of swine, better management practices of swine to prevent exposure of swine to human faeces, inspection of meat and sufficient cooking, and treatment of human taeniasis, and public health education to promote hand hygiene, food safety, sanitation and swine management. (26)

CONCLUSION

Neurocysticercosis is a parasitic disease, predominantly in underdeveloped countries, related to poor sanitation. Most of the population with this disease is asymptomatic, however depending on the site of involvement, inflammatory reaction and numbers of cysticerci, it can be serious and even fatal. With this, new interventionist and educational measures must be created so that there is no such type of contamination of the disease by society.

REFERENCES

1. Bandres JC, White AC Jr, Samo T, Murphy EC, Harris RL. Extraparenchymal neurocysticercosis: report of five cases and review of management. Clin Infect Dis 1992;15:799811.

2. Flisser A, Sarti E, Lightowlers M, Schahtz P. Neuro¬- cysticercosis: regional status epidemiology, impact and control measures in the Americans. Acta Trop. 2003; 87(1):43-51.

3. Welburn SC, Beange I, Ducrotoy MJ, Okello AL. The neglected zoonoses – The case for integrated control and advocacy. Clin Microbiol Infect. 2015; 21(5):433- 43.

4. Del Brutto OH, García HH. Taenia solium Cysticercosis–The lessons of history. J Neurological Sciences. 2015; 359(1-2):392-5.

5. Takayanagui, O. M., & Leite, J. P. (2001). Neurocisticercose. Revista Da Sociedade Brasileira de Medicina Tropical, 34(3), 283–290.

6. Carpio A, Placencia M, Santillan F, Escobar A. A proposal for classification of neurocysticercosis. Canadian Journal of Neurological Sciences 21: 43-47, 1994

7. Machado LR, Nobrega JP, Barros NG, Livramento JA, Bacheschi LA, Spina-Franca A. Computed tomography in neurocysticercosis: a 10-year long evolution analysis of 100 patients with an appraisal of a new classification. Arquivos de Neuropsiquiatria 48: 414- 418, 1990.

8. World Health Organization. (2021). WHO guidelines on management of Taenia solium neurocysticercosis. World Health Organization.

9. Garcia HH, Del Brutto OH. Imaging findings in neurocysticercosis. Acta Trop. 2003; 87(1):71– 8.

10. Arseni C, Samitca DC. Cysticercosis of the brain. British Medical Journal 2: 494-497, 1957.

11. Roman G, Sotelo J, Del Brutto O, Flisser A, Dumas M, Wadia N et al. A proposal to declare neurocysticercosis an international reportable disease. Bull World Health Organ. 2000;78(3):399–406.

12. Del Brutto OH, Salgado P, Lama J, Del Brutto VJ, Campos X, Zambrano M et al. Calcified neurocysticercosis associates with hippocampal atrophy: a population-based study. Am J Trop Med Hyg. 2015;92(1):64–8.

13. Carpio A, Kelvin EA, Bagiella E, Leslie D, Leon P, Andrews H et al. Effects of albendazole treatment on neurocysticercosis: a randomised controlled trial. J Neurol Neurosurg Psychiatry. 2008;79(9):1050–5.

14. Das K, Mondal GP, Banerjee M, Mukherjee BB, Singh OP. Role of antiparasitic therapy for seizures and resolution of lesions in neurocysticercosis patients: an 8 year randomised study. J Clin Neurosci. 2007;14(12):1172–7.

15. Padma MV, Behari M, Misra NK, Ahuja GK. Albendazole in neurocysticercosis. Natl Med J 90 India. 1995;8(6):255–8

16. Garg RK, Karak B, Mohan Kar A. Neuroimaging abnormalities in Indian patients with uncontrolled partial seizures. Seizure. 1998;7(6):497–500.

17. Puri V, Gupta RK. Magnetic resonance imaging evaluation of focal computed tomography abnormality in epilepsy. Epilepsia. 1991;32(4):460–6.

18. Morgado C, Gomes LB, de Campos JG. Neurocysticercosis. An imaging analysis of 35 cases. Acta Med Port. 1994;7(5):269–75.

19. Zee CS, Segall HD, Boswell W, Ahmadi J, Nelson M, Colletti P. MR imaging of neurocysticercosis. J Comput Assist Tomogr. 1988;12(6):927–34.

20. Teitelbaum GP, Otto RJ, Lin M, Watanabe AT, Stull MA, Manz HJ et al. MR imaging of neurocysticercosis. Am J Roentgenol. 1989;153(4):857–66.

21. Jung H, Hurtado M, Medina MT, Sanchez M, Sotelo J. Dexamethasone increases plasma levels of albendazole. J Neurol. 1990;237(5):279–80.

22. Garcia HH, Pretell EJ, Gilman RH, Martinez SM, Moulton LH, Del Brutto OH et al. A trial of antiparasitic treatment to reduce the rate of seizures due to cerebral cysticercosis. N Engl J Med. 2004;350(3):249–58.

23. Del Brutto OH, Sotelo J. Neurocysticercosis: an update. Rev Infect Dis. 1988;10(6):1075– 87.

24. Winkler AS, Richter H. Landscape analysis: management of neurocysticercosis with an emphasis on low- and middle-income countries. Washington (DC): Pan American Health Organization; 2015.

25. Medina MT, Genton P, Montoya MC, Córdova S, Dravet C, Sotelo J. Effect of anticysticercal treatment on the prognosis of epilepsy in neurocysticercosis: a pilot trial. Epilepsia. 1993;34(6):1024–7.

26. Garcia HH, Gonzales I, Lescano AG, Bustos JA, Pretell EJ, Saavedra H et al. Enhanced steroid dosing reduces seizures during antiparasitic treatment for cysticercosis and early after. Epilepsia. 2014;55(9):1452–9.

27. Colli BO, Martelli N, Assirati Jr JA, Machado HR, Salvarani CP, Sassoli VP, Forjaz SV. Cysticercosis of the central nervous system. I. Surgical treatment of cerebral cysticercosis: a 23 years experience in the Hospital das Clinicas of Ribeirao Preto Medical School. Arquivos de Neuropsiquiatria 52: 166-186, 1994

28. Takayanagui OM. [Neurocisticercose. II. Avaliação da terapêutica com praziquantel]. Arquivos de Neuropsiquiatria 48: 11-15, 1990.

29. Takayanagui OM. Albendazole therapy for neurocysticercosis [letter; comment]. Neurology 50: 834-835, 1998.

30. White AC Jr. Neurocysticercosis: updates on epidemiology, pathogenesis, diagnosis, and management. Annu Rev Med. 2000; 51:187-206.

31. Baird RA, Wiebe S, Zunt JR, Halperin JJ, Gronseth G, Roos KL. Evidence-based guideline: Treatment of parenchymal neurocysticercosis: Report of the guideline development subcommittee of the American Academy of Neurology. Neurology. 2013; 80(15):1424–9.

32. Diazgranados JA, Barrios G., Costa JL, Burbano J, Pinzón J. Ivermectina como alternativa terapéutica en neurocisticercosis resistente al tratamiento farmacológico convencional. Revista de Neurología. 2008; 46(11):671-4

33. Working to overcome the global impact of neglected tropical diseases: first WHO report on neglected tropical diseases. Geneva: World Health Organization; 2010.