

# Pearls & Oysters: A rare case of neurotrichinosis with MRI

Christine M. McDonald,  
MD  
Peter Tai, MD, FRCPC,  
ABPN  
Timo Krings, MD, PhD,  
FRCPC(C)

Correspondence to  
Dr. McDonald:  
CMcdonald2@mtsinai.on.ca

## PEARLS

1. The neurologic manifestations of trichinosis are diverse and range from encephalopathy to cerebral, cerebellar, and spinal cord abnormalities.
2. MRI lesions enhance with contrast and are postulated to represent small areas of infarction.
3. Imaging typically normalizes within 1 to 2 months after infection.

## OY-STERS

1. Neurotrichinosis should be considered in patients with marked eosinophilia and unexplained neurologic symptoms.
2. Outcomes may be improved with the administration of steroids and anthelmintic agents during the enteral phase of infection.

**CASE REPORT** A 41-year-old left-handed man from a northern Canadian district presented to a community hospital with a 3-week history of watery diarrhea, a self-resolved erythematous pruritic rash, fever, confusion, and facial swelling. A CT scan of the head showed areas of decreased attenuation bilaterally in the periventricular regions of the frontal lobes. He was offered admission for workup but declined and was sent home with antibiotics. He returned to the hospital 3 days later with new complaints of left face numbness as well as left arm numbness, weakness, and clumsiness. His partner noted that he was sleeping more than usual and had word-finding difficulties. Other symptoms included daily bifrontal headache, “red spots” briefly appearing in his visual field, and ataxia.

Medical history was significant for vasectomy, 20-pack-year smoking history, and headaches. He had no allergies and no regular medications.

On admission, vital signs were within normal limits, and there was no documented fever. Cardiorespiratory and abdominal examination results were normal. There was no facial edema or skin lesions. Cognition was intact. He had a normal cranial nerve

and sensory examination. Strength in the left upper extremity was decreased to 4/5 in all muscle groups. The remainder of the strength examination was normal. Reflexes were present and symmetric. There was a left-sided pronator drift, slowing of finger-to-nose testing, and inability to perform rapid alternating movements with the left arm. Gait was unstable, and the patient used a wheelchair to mobilize in the hospital.

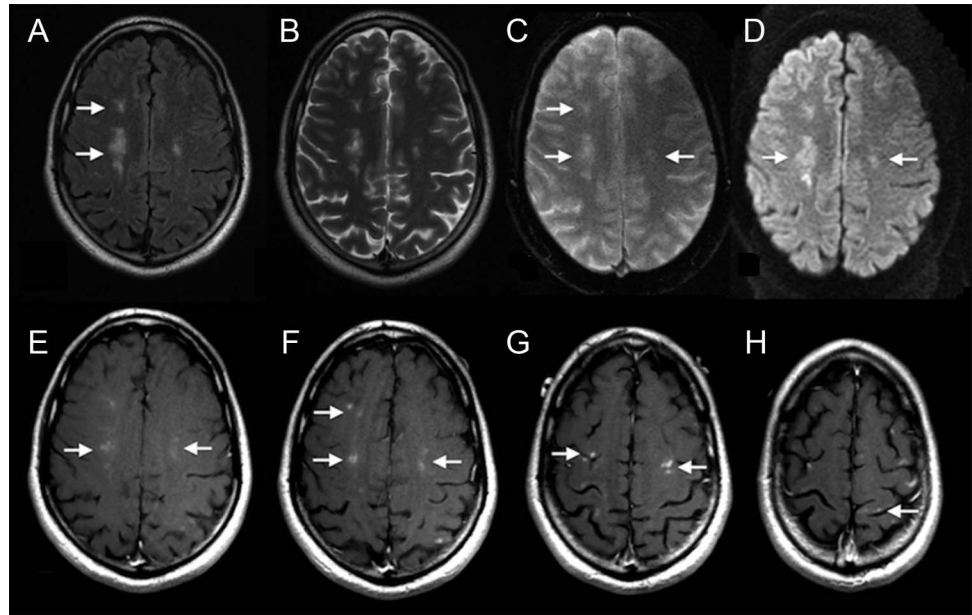
Laboratory tests were remarkable for a leukocyte count of  $15.3 \times 10^9/L$ , an eosinophil count of  $5.8 \times 10^9/L$  (38%), platelets of  $147 \times 10^9/L$ , creatine kinase of 290 U/L, troponin of 3.12  $\mu g/L$ , and erythrocyte sedimentation rate of 34 mm/h. There was no evidence of ST elevation on ECG, and chest x-ray was normal. CSF analysis showed glucose of 57.6 mg/dL, protein 34 mg/dL, erythrocytes  $9.5/mm^3$ , leukocytes  $3.6/mm^3$ , and a negative Gram stain and culture. There were too few leukocytes in the CSF to provide a differential. Blood cultures, stool culture, and stool for ova and parasites were all negative. Rheumatologic markers were negative. Echocardiogram showed normal ventricular and valvular function. An EEG showed slow-wave activity in the left temporal region. There was no epileptiform activity.

Brain MRI (figure) demonstrated multiple bilateral areas of increased signal intensity in both the deep white matter and gray–white matter interface of the frontal and parietal lobes bilaterally, with very few lesions seen in the occipital and temporal lobes and the cerebellum on T2 fluid-attenuated inversion recovery (FLAIR) and T2-weighted scans. Diffusion-weighted scans demonstrated restriction in some of the deep white matter lesions, indicating ischemia. Lesions were slightly hypointense on unenhanced T1 images and there was no pathologic blooming artifact on T2 gradient echo sequences that would have suggested blood products. Following contrast administration, there was an intense rim of enhancement in most lesions. The area of enhancement was smaller than 4 mm in all lesions, with the T2 and FLAIR signal abnormality surrounding the lesions measuring up to 8 mm.

From the Department of Medicine (C.M.M.), Mount Sinai Hospital, Toronto; Department of Medicine (P.T.) and Neuroradiology (T.K.), University of Toronto (C.M.M.); and the Toronto Western Hospital & University Health Network (P.T., T.K.), Canada.

Go to [Neurology.org](http://Neurology.org) for full disclosures. Funding information and disclosures deemed relevant by the authors, if any, are provided at the end of the article.

**Figure** Multimodal MRI of *Trichinella* infection involving the brain



Multimodal MRI including T2 fluid-attenuated inversion recovery (FLAIR)-weighted sequences (A), T2 turbo spin-echo sequences (B), T2 gradient echo sequences (C), and diffusion-weighted imaging scans (D), as well as contrast-enhanced T1-weighted sequences (E-H), demonstrate multiple discrete, nonconfluent foci of T2 and FLAIR hyperintensity in the deep white matter (arrows). There is no abnormal blooming on T2 gradient echo sequences, but restricted diffusion is seen in some of the lesions. Contrast enhancement is visualized in both the deep white matter and multiple cortical/subcortical regions. Images were obtained at approximately 7 weeks in the patient's disease course.

The patient was transferred to our center approximately 7 weeks into his disease course for further neurologic opinion. He complained of persistent headache, myalgias, and difficulty with concentration. A dietary history was elicited, and the patient reported consuming undercooked black bear meat over the preceding 3 months. He was an avid hunter. *Trichinella* serology by complement fixation was reactive with a titer of 1:16. The patient was diagnosed with trichinosis complicated by myocarditis and CNS involvement. He was managed with an infectious disease consultant, and, taking into consideration the improvement of CNS symptoms, stability of MRI findings, and late stage in the course of infection, steroids and anthelmintics were not prescribed. He was discharged with follow-up, and the local public health agency was involved for contact tracing.

**DISCUSSION** Trichinosis is a parasitic infection caused by nematodes of the genus *Trichinella*, with transmission occurring through ingestion of undercooked infected meat. The primary reservoir is pigs, although transmission through game (e.g., wild boar, bear, walrus) is also reported. The parasite is found worldwide, and 66 cases of human infection were reported in the United States between 2002 and 2007.<sup>1</sup> The *Trichinella* life cycle involves an enteral and a parenteral phase. During the enteral phase, encysted larvae are ingested by the host, released by

gastric acid in the stomach, and invade the small intestinal mucosa. The larvae mature and reproduce, releasing newborn larvae into intestinal lymphatics 7 to 10 days after ingestion. The parenteral phase commences with hematogenous spread of larvae to various tissues, including striated muscle. The larvae encyst in muscle tissue and can remain viable for months to years.<sup>2</sup>

Patients with trichinosis may present with diarrhea, fever, periorbital edema, weakness, rash, and myalgias.<sup>3</sup> Bloodwork shows marked eosinophilia. *Trichinella* infection of the CNS, referred to as neurotrichinosis, occurs in as few as 0.2% but as many as 52% of cases and represents serious infection.<sup>4</sup> Neurologic signs and symptoms are nonspecific and range from encephalopathy to hemiparesis and ataxia. For this reason, neurotrichinosis is clinically underrecognized.<sup>5</sup> Cases associated with outbreaks are identified more readily as compared to sporadic infections.<sup>6</sup>

CNS injury is through both direct and indirect mechanisms. Larvae migrate to the brain and cause local destruction, obstruct vasculature, and provoke an inflammatory response.<sup>7</sup> There is no predilection for specific areas of the CNS.<sup>4</sup> Vascular changes, primarily microinfarctions, are thought to be most responsible for neurologic injury and may be mediated by the hyper eosinophilic state.<sup>5,6</sup>

In previous reports, CT imaging of the brain revealed multifocal hypodensities in the cerebral cortex and white matter.<sup>5,6</sup> The MRI appearance of

neurotrichinosis is less frequently described in the literature. Lesions appear hyperintense on T2-weighted images, enhance with gadolinium, and show restricted diffusion on diffusion-weighted imaging.<sup>8</sup> Imaging findings correlate with pathologic findings of cortical and white matter microinfarctions.<sup>6</sup> Findings on imaging typically improve within 1 to 2 months of infection.<sup>4</sup> The CT and MRI findings in our case were consistent with what has been previously described and were suggestive of ischemic injuries of varying ages. MRI findings of neurotrichinosis are nonspecific but argue against alternate diagnoses in our case: the absence of significant edema surrounding the lesions is inconsistent with septic emboli or metastases; the dense enhancement of lesions over a long period of time is inconsistent with ischemic embolic lesions; the absence of abnormal blooming on T2 gradient echo sequences speaks against mycotic aneurysms; and the absence of leptomeningeal enhancement is inconsistent with sarcoidosis or other granulomatous disease.

The diagnosis of neurotrichinosis is based on historical and clinical features. Bloodwork typically reveals hypereosinophilia. The differential diagnosis of hypereosinophilia, defined as eosinophils  $>1.5 \times 10^9/L$ , is broad, but treatable helminth and other parasitic infections must be ruled out. Medication-related eosinophilia, allergic and immunologic diseases, hematologic malignancy, and organ-specific eosinophilic processes (e.g., eosinophilic pneumonia) should be considered.<sup>9</sup> Hypereosinophilia independent of its cause can lead to end-organ damage, including life-threatening cardiomyopathy.<sup>5,9</sup> Serologic tests are helpful in the diagnosis of trichinosis.<sup>2</sup> A definitive diagnosis can be achieved through muscle biopsy, although this is rarely necessary. Low eosinophil counts or a rapid fall in the count seem to be associated with poor outcomes.<sup>2</sup>

Albendazole or mebendazole is the standard treatment in neurotrichinosis. Improvement of symptoms with the use of anthelmintics vs placebo has been shown in one small randomized controlled trial.<sup>10</sup> Treatment is most effective during the enteral phase, as migration and muscular invasion can be attenuated. The effect of treating trichinosis in its later stages is not well known and likely less successful once the parasite has encysted in muscle tissue.<sup>2</sup> High-dose corticosteroids are recommended in addition to anthelmintics for patients with severe infection to suppress vascular damage related to eosinophilia.<sup>4</sup>

The prognosis of patients with neurologic complications of trichinosis is variable, and, despite treatment,

some patients have chronic sequelae. Mortality is as high as 5% when the CNS is involved.<sup>2</sup> For this reason, patients with neurotrichinosis should be hospitalized for observation and investigated for multisystem involvement. Our patient made a near-full neurologic recovery in 2 months, with his primary residual deficits being a very mild encephalopathy and headache.

## AUTHOR CONTRIBUTIONS

Dr. McDonald participated in patient care and wrote and revised the manuscript. Dr. Tai participated in patient care and provided revisions to the manuscript. Dr. Krings provided MRI interpretation, the figure, and revisions to the manuscript.

## STUDY FUNDING

No targeted funding reported.

## DISCLOSURE

C. McDonald reports no disclosures. P. Tai is a staff neurologist at University Health Network in Toronto and has received funding as a site investigator from Upsher-Smith Laboratories in an unrelated randomized double-blind placebo-controlled study of the safety and efficacy of intranasal midazolam in outpatient treatment of patients with seizure clusters. T. Krings reports no disclosures. Go to Neurology.org for full disclosures.

## REFERENCES

1. Kennedy ED, Hall RL, Montgomery SP, Pyburn DG, Jones JL. Trichinellosis surveillance: United States, 2002–2007. *MMWR Surveill Summ* 2009;58:1–7.
2. Gottstein B, Pozio E, Nockler K. Epidemiology, diagnosis, treatment, and control of trichinellosis. *Clin Microbiol Rev* 2009;22:127–145.
3. Hall RL, Lindsay A, Hammond C, et al. Outbreak of human trichinellosis in Northern California caused by *Trichinella murrelli*. *Am J Trop Med Hyg* 2012;87:297–302.
4. Neghina R, Neghina AM, Marincu I, Iacobiciu I. Reviews on trichinellosis (II): neurological involvement. *Foodborne Pathog Dis* 2011;8:579–585.
5. Fourestie V, Douceron H, Brugieres P, Ancelle T, Lejonc JL, Gherardi RK. Neurotrichinosis: a cerebrovascular disease associated with myocardial injury and hypereosinophilia. *Brain* 1993;116:603–616.
6. Ellrodt A, Halfon P, Le Bras P, et al. Multifocal central nervous system lesions in three patients with trichinosis. *Arch Neurol* 1987;44:432–434.
7. Gay T, Pankey GA, Beckman EN, Washington P, Bell KA. Fatal CNS trichinosis. *JAMA* 1982;247:1024–1025.
8. Gelal F, Kumral E, Dirim Vidinli B, Erdogan D, Yucel K, Erdogan N. Diffusion-weighted and conventional MR imaging in neurotrichinosis. *Acta Radiol* 2005;46:196–199.
9. Roufosse F, Weller PF. Practical approach to the patient with hypereosinophilia. *J Allergy Clin Immunol* 2010;126:39–44.
10. Watt G, Saisorn S, Jongsakul K, Sakolvaree Y, Chaicumpa W. Blinded, placebo-controlled trial of antiparasitic drugs for trichinosis myositis. *J Infect Dis* 2000;182:371–374.

# Neurology®

## Pearls & Oy-sters: A rare case of neurotrichinosis with MRI

Christine M. McDonald, Peter Tai and Timo Krings

*Neurology* 2014;82:e30-e32

DOI 10.1212/WNL.0000000000000051

**This information is current as of January 27, 2014**

<b>Updated Information &amp; Services</b>	including high resolution figures, can be found at: <a href="http://n.neurology.org/content/82/4/e30.full">http://n.neurology.org/content/82/4/e30.full</a>
<b>References</b>	This article cites 10 articles, 2 of which you can access for free at: <a href="http://n.neurology.org/content/82/4/e30.full#ref-list-1">http://n.neurology.org/content/82/4/e30.full#ref-list-1</a>
<b>Subspecialty Collections</b>	This article, along with others on similar topics, appears in the following collection(s): <b>Cardiac; see Cerebrovascular Disease/Cardiac</b> <a href="http://n.neurology.org/cgi/collection/cardiac_see_cerebrovascular_disease-cardiac">http://n.neurology.org/cgi/collection/cardiac_see_cerebrovascular_disease-cardiac</a> <b>Clinical neurology history</b> <a href="http://n.neurology.org/cgi/collection/clinical_neurology_history">http://n.neurology.org/cgi/collection/clinical_neurology_history</a> <b>MRI</b> <a href="http://n.neurology.org/cgi/collection/mri">http://n.neurology.org/cgi/collection/mri</a>
<b>Permissions &amp; Licensing</b>	Information about reproducing this article in parts (figures, tables) or in its entirety can be found online at: <a href="http://www.neurology.org/about/about_the_journal#permissions">http://www.neurology.org/about/about_the_journal#permissions</a>
<b>Reprints</b>	Information about ordering reprints can be found online: <a href="http://n.neurology.org/subscribers/advertise">http://n.neurology.org/subscribers/advertise</a>

*Neurology*® is the official journal of the American Academy of Neurology. Published continuously since 1951, it is now a weekly with 48 issues per year. Copyright © 2014 American Academy of Neurology. All rights reserved. Print ISSN: 0028-3878. Online ISSN: 1526-632X.

